Behavioural Adherence in the Treatments of Disorders of Sleep and Wakefulness- A Biopsychosocial approach

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Abstract

Obstructive sleep apnoea (OSA) and insomnia are the two most prevalent sleep disorders. Their respective treatments Continuous Positive Airway Pressure (CPAP) and Cognitive Behaviour Therapy for insomnia (CBT-I), are effective, but at the same time challenging. It is this challenge that may translate to poor adherence, which ultimately leads to a reduction in treatment effectiveness. The evaluation of these treatments should not fall short of understanding effectiveness by only considering efficacy; the effort to establish what influences adherence makes up a large part of that goal. The aim of this thesis is to contribute to the literature by adopting a biopsychosocial approach (BPS). That is, the consideration of biomedical, psychological and social factors and how they interact to influence behaviour. The implications for both CPAP and CBT-I adherence literature were tested in the context of four experimental studies.

Semi-structured interviews were conducted with 11 CPAP users, with 5 individuals completing the three required interviews prior to, at 1 week and 3 months after treatment initiation. The core themes emerging from a thematic analysis were 'internal conflict around acceptance and adherence', 'integration of CPAP into life' and 'motivators and resources for CPAP use'. The interviews with 11 individuals having completed a CBT-I program revealed three important issues: 'Making sense of CBT-I', 'Ongoing evaluation of components' and 'Obstacles to implementation'. Both studies reveal potential psychological and social factors contributing to adherence to CPAP and CBT-I, which need to be considered in a BPS framework.

A patient-level meta-analysis of three randomised placebo-control studies showed that the relationship between CPAP adherence and improvements in daytime sleepiness was caused by both physiological (high use of real CPAP reduced sleepiness more than high use of placebo and more than low use of real CPAP) and psychological effects (high use of placebo was superior to low use of placebo), possibly as a result of an expectation of benefit. The results support the importance of considering both biomedical and psychosocial factors and their interactive effects on adherence.

The translation of the BPS approach to clinical practice will be facilitated by the development of brief, reliable and valid measures to assess psychological

and social variables in addition to the existing biomedical tools. The Stage of Change Scale for Insomnia (SOCSI) assessing components of the transtheoretical model (stage of change, self-efficacy, decisional balance and processes of change), was constructed and cognitively pre-tested in 13 individuals completing CBT-I. The reliability and validity of this comprehensible scale was subsequently examined in the context of a sleep restriction trial. Insomnia-related symptoms at post-treatment and follow-up, which were significantly different from baseline in the 27 individuals with insomnia, were associated with actigraph-determined adherence to the agreed bed window. The SOCSI was deemed a valid tool with participants in the self-identified action/maintenance stage revealing significantly better adherence, higher motivation and self-efficacy than those in the contemplation and preparation stage. Test-retest reliability of the SOCSI was excellent and the content analysis of open-box responses revealed information for further validation of decisional balance and processes of change scales.

This thesis provides novel information about the variables that influence adherence to CPAP and CBT-I. It distinguishes itself from previous efforts by acknowledging the need for the adoption of a BPS framework. This approach is necessary to successfully advancing not only the CPAP and CBT-I adherence literature individually, but potentially the adherence field in general.

Table of Content

List of Figures

List of Tables

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I would like to dedicate this thesis to my sister Alys Crawford. Alys, you know why. Ich liebe dich!

Author's Declaration

I declare that, except when explicit reference is made to the contribution of others, that this thesis is the result of my own work and has not been submitted for any other degree at the University of Glasgow or any other institution.

Signature: 

Print Name: Megan Crawford

Abbreviations

AASM= American Association of Sleep Medicine

APA= American Psychiatric Association

AHI= Apnea Hypopnea Index

BPS= Biopsychosocial

CBT= Cognitive Behavioural Therapy

CBT-I= Cognitive Behavioural Therapy for Insomnia

CPAP= Continuous Positive Airway Pressure

DBAS= Dysfunctional Beliefs and Attitudes about Sleep

EOG= Electrooculography

EEG= Electroencephalogram

EMA= Early morning awakening

EMG= Electromyography

HADS= Hospital Anxiety and Depression Scale

II= Idiopathic Insomnia

ISI= Insomnia Severity Index

MAD= Mandibular Advancement Device

MSLT=Multiple Sleep Latency Test

ODI= Oxygen Desaturation Index

OSA= Obstructive Sleep Apnea

RCT= Randomised Control Trial

RDI= Respiratory Disturbance Index

PI= Paradoxical Insomnia

PSQI= Pittsburgh Sleep Quality Index

PSG= Polysomnography

PPI= Psychophysiological Insomnia

SE= Sleep efficiency

SCT= Stimulus Control Therapy

SDB= Sleep Disordered Breathing

SOCSI= Stage of Change Scale for Insomnia

SOC= Stage of Change

SOL= Sleep Onset Latency

SRT= Sleep Restriction Therapy

TIB= Time in Bed

TST= Total Sleep Time

WASO= Wake after sleep onset

Overview

Thesis Overview

This section aims to provide a brief chapter-by-chapter overview of the thesis. This Ph.D. was completed as a Cotutelle Agreement between the Universities of Glasgow (United Kingdom) and Sydney (Australia). The sleep research facilities at the respective universities have expertise in both obstructive sleep apnoea and insomnia. Consequently, this thesis has considered these two clinical populations and presented them in this thesis as separate disorders.

Chapter 1- Introduction

This chapter provides a brief overview of the diagnosis, pathophysiology and treatments of both sleep disorders, which is relatively brief. With the main focus of this thesis directed towards adherence to treatments, the large part of this chapter presents the measurement and predictors of and interventions to improve adherence.

Chapter 2-Literature Review

Chapter 2 is a narrative review of the CPAP adherence literature and provides the basis for the thesis aims: a biopsychosocial (BPS) approach to adherence. The CPAP literature was chosen as the focus, merely because of the extensive literature available in this area and it provides a good example of where biomedical and psychosocial models of adherence diverge. However the research and clinical implications can be transferred to the CBT-I adherence literature as discussed where appropriate. At the time of thesis submission, this review was under peer-review in the journal Sleep Medicine Reviews. The finished article (with slight adaptations for the thesis) is presented in chapter 2 and thus some information might slightly overlap with the introduction in chapter 1. The thesis aims are presented at the end of this chapter.

Chapter 3-6 (Experimental Chapters)

The experimental chapters have the purpose of reaching the broad aims of the thesis, which stem from the overarching aim to adopt a BPS approach to adherence described in chapter 2. The qualitative studies outlined in chapter 3

and 4 offer information particularly about psychological and social factors that influence both CPAP and CBT-I in addition to biomedical factors. The patient-level meta-analysis in chapter 5 provides initial support for integrating biomedical, psychological and social aspects into a holistic model. Finally, chapter 6 describes the development and validation of a scale to assess the psychological construct of readiness to change, contributing to building a database of brief scales that will facilitate the identification of bio-, psycho-, social variables that predict adherence to treatment for the disorders of interest. At the time of submission, Chapter 5 was published in the journal Thorax. This paper has been included in this thesis as complete work and thus any references to this chapter are to the publication (Crawford et al. 2012).(1) Slight adaptations have been made for the inclusion in the thesis (e.g., supplementary material are included in the appendix). The methodology and result of each chapter are critically evaluated in each individual chapter discussions; further directions for research and implications for clinical practice are also provided here. As the work in this thesis is pragmatic in its nature the discussion for each experimental chapter will not include separate sections for research and clinical practice, but these are interwoven in each discussion. The experimental chapters are then assessed in context with the thesis aims in the overarching discussion.

Chapter 7 Overarching Discussion

This section aims to merge the information provided in each experimental chapter within the context of the thesis aims. Results particularly relevant to consider within the biopsychosocial framework are discussed in this section, with further information on future directions and strengths and limitations of the thesis as an overall body of work.

Chapter 1- Introduction to Disorders of Sleep and Wakefulness

1.1 Obstructive Sleep Apnoea

The International Classifications of Sleep Disorders-2 diagnostic nosology (ICSD-2)(2) classifies sleep related breathing disorders (SRBD) into four distinct categories: Central Sleep Apnea Syndromes, Obstructive Sleep Apnea Syndromes, Sleep Related Hypoventilation/Hypoxemic Syndromes, and Other Sleep Related Breathing Disorders. The underlying characteristic is an abnormality in breathing that solely occurs in or is even exacerbated by the sleep state. The most common condition- obstructive sleep apnoea in adults (from hereon referred to as OSA)- is given exclusive attention in this thesis, thus the introduction will be reduced to the diagnostic and pathophysiological descriptions and treatments of this disorder only.

1.1.1 Diagnostic Classification

OSA is characterised by the obstruction of the upper airway during sleep that leads to complete cessation (apnoea) or reduction (hypopnoea) of airflow. This occurs irrespective of continued ventilatory effort. Before termination, most often through arousals, these events lead to a decrease in blood oxygen saturation and associated increase in carbon dioxide levels during longer events. These effects (disrupted sleep and hypoxemia) are identified as the possible causal factor for the daytime sleepiness and resulting impairment witnessed in these individuals.(3)

According to the ICSD-2 the diagnosis of obstructive sleep disordered breathing is reliant on the clinical (e.g. daytime sleepiness, breathing interruptions or snoring and witnessed gasping for air) and, or just polysomnographic (respiratory events) characteristics.(2) Table 1 highlights the diagnostic criteria outlined by the ICSD-2.

Table 1: Diagnostic criteria for OSA according to the ICSD-2

ICSD-2 criteria for obstructive sleep apnoea (verbatim from 2):
A,B and D or C and D satisfy the criteria:
A. At least one of the following applies:
i) The patient complains of unintentional sleep episodes during wakefulness, daytime sleepiness, unrefreshing sleep, fatigue or insomnia.
ii) The patient wakes with breath holding, gasping, or choking.
iii) The bed partner reports loud snoring, breathing interruptions, or both during the patient's sleep
B. Polysomnographic recording shows the following:
i) Five or more scoreable respiratory events (i.e. apneas, hypopneas, or RERAs) per hour of sleep
ii) Evidence of respiratory effort during all or a portion of each respiratory event (In the case of a RERA, this is best seen with the use of esophageal manometry)
OR:
C. Polysomnographic recording shows the following
i) Fifteen or more scoreable respiratory events (i.e. apneas, hypopneas or RERAs) per hour of sleep
ii) Evidence of respiratory effort during all or a portion of each respiratory event (In the case of a RERAs, this is best seen with the use of esophageal manometry).
D. The disorder is not better explained by another current sleep disorder, medical or neurological disorder medication use, or substance use disorder.

The AASM has defined an apnoea, objectively determined by polysomnography (PSG), as a complete pause in airflow (more than 90% reduction from baseline) for at least 10 seconds. An airflow reduction of at least 30% (alternative 50%) with an associated minimum 4% decrease in oxygen saturation (alternative 3% or associated arousal), lasting for at least 10 seconds is classified as a hypopnoea. Lastly, the respiratory effort-related arousal event (RERAs) are episodes that last at least 10 seconds. Increased respiratory effort follows these events eventually leading to an arousal from sleep. The RERAs do not meet criteria for apnoea or hypopnoea and are therefore included in the respiratory disturbance index (RDI), but not to Apnoea/Hypopnoea Index (AHI) (4). Apart from the AHI and RDI, the oxygen desaturation index (ODI) may be used as a marker of severity. The ODI measures the amount of times per hour the oxygen saturation levels fall >3 or 4% below the baseline value.(4)

The AHI, the number of apnoeas and hypopnoeas per hour of recorded sleep, is most commonly used, and cut-offs for severity are as followed(3):

mild OSA= AHI between 5 and 15

moderate OSA= AHI between 15 and 30

severe OSA= AHI greater than 30

Obstructive sleep apnoea can be differentially diagnosed from Central Sleep Apnoea, the latter condition being associated with an absence of respiratory effort. However events in the OSA patient may also be central or even mixed in nature, which is defined by central followed by obstructive characteristics.(3)

1.1.2 Diagnosis of OSA

As alluded to above, the main clinical features of OSA are snoring, witnessed apnoeic events and daytime sleepiness (non-specific symptom of OSA and often measured with the Epworth Sleepiness Scale, ESS(5)). Clinical risk factors that OSA patients might present with in clinical practice are obesity,(6) or more likely visceral fat(7), increased neck circumference,(8) older age(9) and male gender.(10) Certain craniofacial features (e.g. size and position of the mandible and maxilla(11, 12)) and upper airway abnormalities (e.g. narrow posterior pharynx, airway length(13, 14)) are often related to the prevalence of OSA, or at least a more collapse prone airway. Consequences of OSA, as outlined below, such as impaired cognition and performance(15) low mood/depression(16) and sexual/erectile dysfunction(17) may also be signals for the practitioner at the first point of contact.

Prediction models based on these clinical features are however not sufficient for diagnosis, and the “gold standard” clinical pathway for the diagnosis of OSA is through an in-lab attended polysomnogram (PSG).(18) A full PSG includes a variety of measures that obtain information about electrical activity of the brain, muscle and eye movement, respiration, oxygen saturation in the blood, respiratory effort and cardiac rhythm, usually measured in 30-second epochs. Electrical brain activity, as measured by the electroencephalogram (EEG) is a marker of objective sleep and provides partial information about sleep stages. It is not within the boundaries of this thesis to give an extensive description of sleep architecture, but it is sufficient to say that sleep occurs in stages, cycling in roughly 90 minute episodes between lighter (higher wave frequency) stages 1 and 2, to deeper (lower wave frequency) stages 3, 4 and Rapid Eye Movement Sleep (REM). Stages 1-4, also termed Non-REM sleep, are most prevalent during the first part of the night and dissipate, whilst REM sleep increases in length and frequency towards morning.(19) The two other channels that help to define stages of sleep are electromyography

(EMG), which measures muscle activity and electrooculography (EOG) which records eye rotation; output from these channels can measure the loss of muscle tone and the rolling eye movement, respectively, both of which are characteristic of REM sleep.

However what is essential to the diagnosis of OSA are the recordings of respiratory events (airflow), and blood oxygen levels, respectively measured by a nasal cannula pressure transducer (superior to a thermistor(20)) and pulse oximetry. The former measurement is needed to record a reduction in airflow, which detects both apnoeas and hypopnoeas, the latter is required for the identification of hypopnoeic events. To distinguish between obstructive and central events thoracic and abdomen movement are recorded as a marker of respiratory effort. Recordings not essential for the diagnosis of OSA, but also included in routine PSG recordings are body position, snoring intensity, cardiac rhythm (electrocardiogram (ECG)) and EMG of the anterior tibialis muscle in order to rule out periodic limb movement disorder (PLMD).

Modifications to the full night in-lab diagnosis may include the split night, unattended portable monitoring (PM) and limited study (excluding EEG, EOG and EMG). PM and limited study can be provided in the patients home. Pulse oximetry as a single channel is not recommended for the diagnosis of OSA, but may be useful for screening purposes.(3) The second half of a split night study is allocated for titration of the pressure delivered during Continuous Positive Airway Pressure (CPAP) treatment, described below. This type of diagnosis, along with PM and *attended* limited studies, is appropriate in selected patients, where certainty of moderate to severe OSA is high.(18, 21)

1.1.3 Pathophysiology

A simplistic view of the complex pathophysiology of OSA(22) is that physiological and situational changes during sleep lead to an imbalance of forces that normally ensure the patency of the airway during wakefulness. Figure 1 is a schematic representation of the forces during wakefulness. During normal respiration whilst awake, the air flows unobstructed through the upper respiratory tract (nasal cavity, pharynx, larynx), through to the lower respiratory tract (trachea, primary bronchi and finally the lungs). To allow for the functions of the upper airway, i.e., speech ventilation and deglutition- this area must rely on both muscle activity and collapsibility.

During inspiration a negative pressure develops, as a result of basic physical principles. Alongside external pressures to the airway (e.g. fat deposition and a small mandible), these negative pressure forces promote the collapse of the airway during inspiration. Counteracting forces (activation of pharyngeal dilator muscles and increase in lung volume) ensure airway patency. The interaction of these forces enables continuous un-obstructive airflow during wakefulness; however changes associated with the sleep state disrupt the balance between these factors, leading to an increased risk of airway collapse. For example a change associated with muscle atonia during REM sleep leads to the reduced activation of the pharyngeal dilator muscle. Supine body position during sleep can also contribute to airway collapse as a result of gravitational forces. Why this partial or full obstruction only occurs in certain individuals is still debated, however it is likely a combination of abnormal anatomy and neuromuscular functioning.(23, 24)

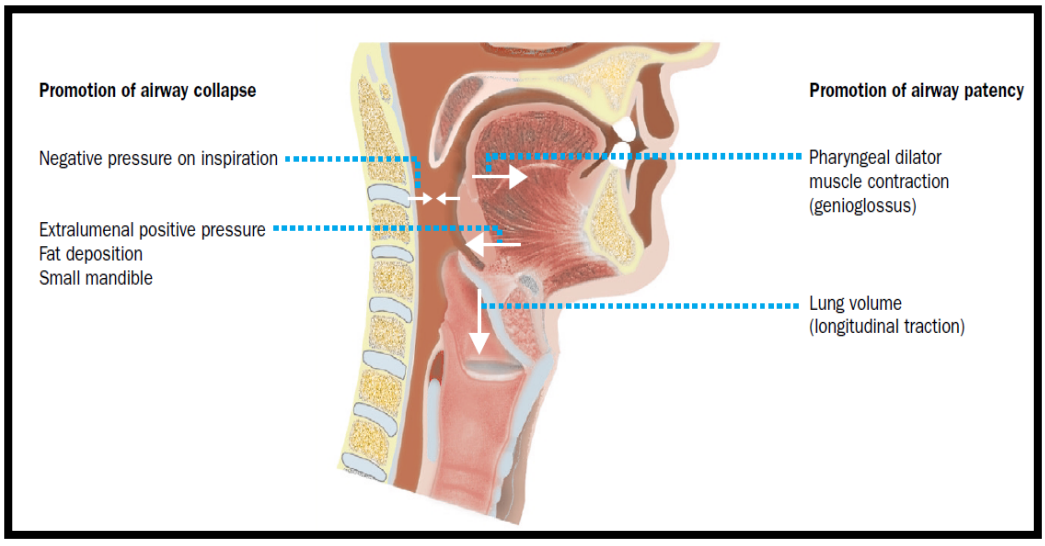


Figure 1: The upper airway during wakefulness
Source: Malhotra & White, Lancet 2002(22)

1.1.4 Prevalence

Obstructive Sleep Apnoea is the second most common sleep disorder following insomnia (see 1.2). Rates can vary extensively depending on the diagnostic criteria used.(3) In 1993 Young derived the prevalence of OSA from the Wisconsin Sleep Study Cohort in the US.(10) Using an AHI cut-off of 5 events per hour, they reported rates of 9 and 24% for women and men respectively. However, when daytime sleepiness was also considered, rates dropped to 2% for women and 4% for men and similar rates have been reported in Australia(25). Reported prevalence rates in the UK are lower, between 1-2%,(26-28) which may

relate to an under-diagnosis of OSA in the UK.(26) Prevalence rates and severity of OSA tend to be higher in non-white minority groups, however not after controlling for differences in body habitus.(29-31)

1.1.5 Treatment of OSA

Treatment of obstructive sleep apnoea can largely be divided into non-implantable mechanical devices (NIMDs), surgery, pharmacological and conservative treatments. One of these NIMDs, Continuous Positive Airway Pressure (CPAP)(32) has been the treatment of choice until recently for the amelioration of apnoeaic, hypopnoeic and RERAs during sleep in all severity ranges.(33)

1.1.5.1 Continuous Positive Airway Pressure

CPAP delivers a fixed pressure of compressed air, creating a pneumatic splint to keep open the airway, thus allowing normal breathing and uninterrupted sleep.(32) Pressure is generated through a machine and delivered through a hose attached to an interface that is sealed closely to the patients face. Interfaces can vary from full head, face or nasal masks or nasal prongs, fitting just over the nostrils. The delivered pressure is individually and manually titrated for each patient. A modification of the in-lab titration with the CPAP machine has evolved through the introduction of the AutoCPAP machine, which automatically detects the occurrence of an event breath-by-breath and increases the pressure to a pre-set maximum from a pre-set minimum. Thus variations of the titration include AutoCPAP unattended at home/ward or attended in the lab, as well as split-night studies as outlined above. Practice parameters indicate the use of AutoCPAP for in-lab titration in moderate or severe cases, and at home only in patients without co-morbidities.(34) AutoCPAP is also used as long-term treatment when preferred over CPAP, or the latter is intolerable.(34) A further modification of pressure delivery is BiPAP, which provides a higher predefined inspiratory than expiratory pressure. Lastly, expiratory pressure relief is a modification to CPAP (also called CFlex), which delivers a slightly lower expiratory level to lessen the feeling of breathing against pressure.

The efficacy of CPAP was apparent in Sullivan's initial presentation of CPAP, where the re-occurrence of obstructive events was witnessed by simply switching off the pressure in all 5 patients.(32) There is now considerable

evidence documenting the efficacy of CPAP to reduce OSA severity(35, 36) and establishing its cost-effectiveness above placebo/conservative treatment.(37) CPAP can reduce the consequences of OSA, such as daytime sleepiness and improve cardiovascular health and driving performance, which is outlined in detail below.

CPAP does not present a cure, but rather a crutch: one night without CPAP can witness the return of nightly and daily symptoms.(38) Adherence to CPAP is generally poor (see below) and some have argued this reduces the potential benefit ('effective AHI')(39, 40) leaving less optimal, but less intrusive, treatments comparable to CPAP. The AASM describes that alternative treatment options may be suitable depending on factors such as treatment preference or anatomical features.(33) Alternatives to CPAP include conservative treatments (weight loss, positional therapy), oral appliances, and surgery and will briefly be reviewed.

1.1.5.2 Other Non-Implantable Medical Devices

Oral appliances (OAs) provide another form of non-implantable mechanical device. These appliances increase the size of the airway using two broad functions; jaw repositioning and tongue retention. Recently updated practice parameters recommend the use of OAs in mild to moderate individuals unable or unwilling to tolerate CPAP, however the use of these devices for severe cases is not indicated at present.(41) In a comprehensive meta-analysis, the use of OAs successfully treated (AHI<5/hr) 35% of patients across randomised placebo controlled cross-over studies. Comparing these types of devices with CPAP (75%), OAs (42%) were more similar to surgery (30%) in percentage of reduction of AHI. Perhaps unsurprisingly, OAs were preferred by patients in the majority of studies, which might have resulted in relatively good adherence rates; 56–68% of patients were still using CPAP after 33 months.(42) Objective use with these devices is still to be established.

There is little evidence for the efficacy of nasal dilators, but this technique might be appropriate for snoring relief.(43, 44) A novel technique making use of a two-wave valve at the nostrils providing positive back-pressure from the individual's own expiratory pressure, was introduced after a pilot study in 2008.(45) Testing this device in 229 OSA patients in a randomised control trial (RCT) revealed significant effects, however the treatment did not reduce the

AHI below 5/hr.(39). Subjective adherence however seems to be excellent, with patients reporting all night use of the device on most nights(46, 47)), which, like OAs, might render it comparable to the poorly adhered to CPAP device.

1.1.5.3 Surgical Management of OSA

Surgery to alleviate symptoms of OSA can occur at the nasal passage (e.g. nasal reconstruction) or the upper airway (e.g. Uvulopalotopharyngoplasty, tracheotomy), or aims to achieve skeletal/geniglossus advancement. A Cochrane review evaluating these surgical options concluded there were too few studies to establish the efficacy of these treatments(48) Removing the obstruction through surgery (e.g. through tonsillectomy, septoplasty) has been hypothesised to improve acceptance and adherence; some studies have shown lowered pressure requirements(49, 50) and increased adherence(51) post surgery.

1.1.5.4 Conservative Treatments

Conservative treatments include lifestyle changes targeting areas that are known risk factors, which exacerbating the frequency of events. These may include excess weight, alcohol, smoking and sleep deprivation. Because of the known association between obesity and OSA, weight loss is most often suggested in conjunction with more effective treatments such as CPAP.(52) The difficulty with this option is the reduced motivation to loose weight in these sleepy individuals, as well as the lack of applicability to the non-obese OSA population,. Likewise, as body position exacerbates the frequency of events, positional therapy (PT, avoidance of the supine position) is often considered as a conservative option. An early study questioned the effectiveness of PT in patients with non position-dependent OSA.(53)

1.1.6 Consequences of OSA and subsequent improvement with CPAP

Figure 2 is a schematic representation of how the obstructive events lead to the known neurocognitive and cardiovascular consequences of the disorder. The termination of the apnoea is often preceded by an arousal, which leads to sleep fragmentation and activation of the sympathetic nervous system. The former explains the pathway to the neurocognitive sequelea, whilst the latter leads to a strain on the cardiovascular system. Fortunately, there is evidence to show that

CPAP is effective in preventing some of these outcomes. Thus, the subsequent sections are devoted to exploring the main consequences of OSA and how effective CPAP use is protective against this sequelae.

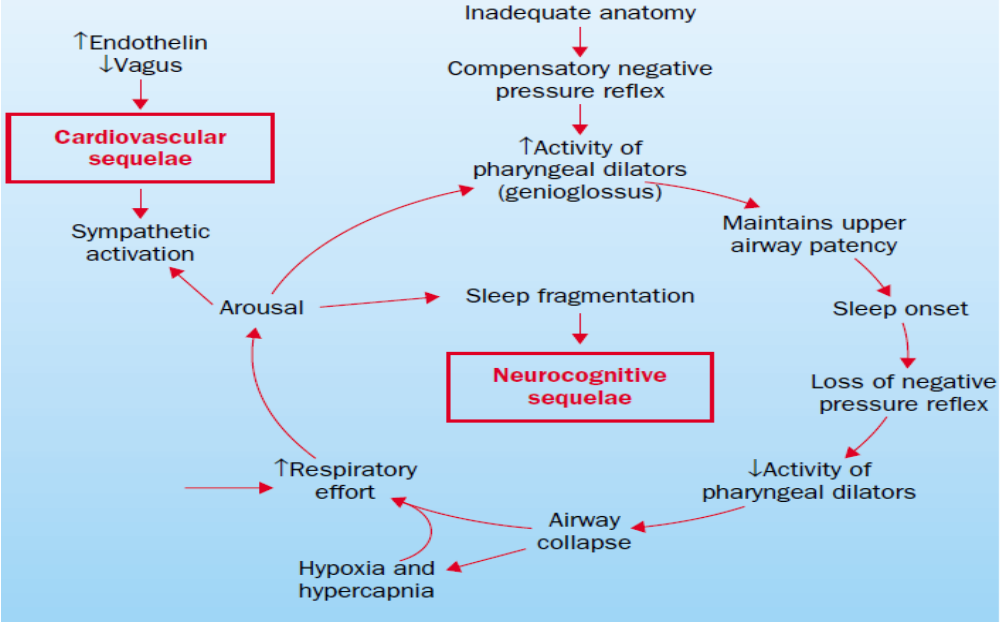


Figure 2: The neurocognitive and cardiovascular consequences of OSA
Source: Malhotra & White, Lancet 2002(22)

1.1.6.1 Daytime Sleepiness

Daytime sleepiness is one of the core symptoms of OSA as a result of the continued arousals from sleep and is often a presenting symptom. In Young's epidemiological evaluation, 23% of women and 16% of men with OSA also reported sleepiness on 2 or more days per week. The authors note these subjective reports are likely underestimating the true hypersomnolence rates,(10) particularly in individuals with mild OSA who are asymptomatic.(54) Recent meta-analyses have reported the effectiveness of CPAP improving both subjective sleepiness and objective sleepiness as measured with the Multiple Sleep Latency Test (measures the propensity to fall asleep) and Maintenance Wakefulness Test (measures the ability to stay awake).(55, 56) A number of studies have indicated this relationship manifests itself in a dose-dependent manor, where increased use of CPAP is associated with increased improvements in sleepiness.(1, 57-59) However both Weaver et al. and Antic et al. report a moderate proportion (10-50%) does not achieve normal values for sleepiness related measures despite adequate use. Further research is necessary to understand this paradox.

1.1.6.2 Cardiovascular Health

A variety of cardiovascular outcomes have been linked to OSA, which includes hypertension, and cardiovascular disease such as stroke. After an initial debate, OSA is now considered an established independent risk factor for hypertension,(60) in sleep clinic patients(61, 62) and in both cross-sectional epidemiological(63-65) and longitudinal studies.(66) A recent longitudinal study, however revealed no relationship in 1,180 patients with OSA after controlling for age, gender, BMI, neck circumference and lifestyle factors such as fitness levels, alcohol and coffee consumption. The authors call for further longitudinal studies to confirm the causal relationship between hypertension and OSA.(67) Support for the link between OSA and hypertension may come from studies showing improvements in blood pressure with CPAP treatment. Two recent meta-analysis, however present contradicting results on the effects of CPAP on this measure.(68, 69) The meta-analysis reporting no significant difference with CPAP use, however do not exclude the effectiveness in selected patient e.g. those with substantial hypertension.(68)

Similarly OSA has been linked to stroke, heart failure and an overall measure of cardiovascular disease/death, with CPAP acting as a protective against the incidence of these events.(70-75)

1.1.6.3 Cognitive Functioning and Performance

As outlined in Figure 2, sleep fragmentation caused by repeated arousals can lead to a wide range of deficits in neurocognitive functioning. This may include attention, memory and executive functioning and performance related quality of life. Whether these deficits are attributable to the sleep disruption caused by the arousals or the intermittent hypoxemia remains unanswered.(15) Furthermore, the effect of CPAP treatment on these domains is still unclear. For example, irrespective of a sound study design, large sample size (1105 OSA patients) and assessing major neurocognitive domains, the delivery of CPAP only produced marked improvements in executive functioning over sham CPAP ($p = 0.0074$) at 2- months, but not attention and psychomotor function, or learning and memory. The authors note that these results along with other negative findings in the literature are suggestive of a complex interaction between OSA and neurocognitive sequelae, with a mediating effect of individual differences in resilience towards sleep deprivation.(76)

One particular concern is road traffic safety. The increased risk of automobile accidents may be two-fold, resulting from lapses in attention through increased sleepiness and cognitive/perceptual difficulties.(77) Two meta-analyses have recently shown a decrease in automobile accidence both on the road and in a driving simulator(77, 78) and a further indicated that when motor vehicle accidents were taken into account, CPAP represents a cost-effective option for the treatment of OSA.(79)

1.1.6.4 Erectile and sexual functioning

Erectile and sexual (E/S) dysfunction as a potential consequence of OSA are largely under researched and overlooked in clinical practice. A study on 401 male OSA patients found a strong relationship between E/S dysfunction and OSA (AHI>5/hr), irrespective of known associates of E/S dysfunction, such as age or obesity. The authors concluded that hypoxemia likely explains the high prevalence of erectile and sexual dysfunction in this population.(17) When treating OSA with CPAP, marked improvements can be witnessed in this area, as highlighted by a paper presented at the 2012 Associated Professional Sleep Societies Conference in Boston.(80) Interestingly after 1, 3 and 6 months of CPAP use, improvements were documented in both those that did and did not report E/S dysfunction at baseline. In the former group there was a relationship between CPAP use and improvement, however the authors concluded E/S dysfunction did not completely resolve, with only 41% of adequate users reaching normal values on the erectile functioning scale.

1.1.6.5 Mortality

Perhaps unsurprisingly, with the cardiovascular sequelae outlined above, moderate to severe OSA has been linked to a four to six-fold increased risk of mortality, irrespective of factors such as age, diabetes or high cholesterol.(81, 82) Fortunately, CPAP has been shown to increase rates of survival in selected patients, such as stroke patients(83) females(84), and the elderly(85). To date, longitudinal community-based samples have not been completed.

1.1.7 Summary

Despite being the treatment of choice for OSA - a condition associated with a number of health and quality of life related consequences -CPAP is intrusive to the patient’s life, and perhaps unsurprisingly, associated with poor adherence.

In order to prevent the negative sequelae outlined above, it is important to understand what influences adherence and which interventions are most effective in the task of improving CPAP use; this will be discussed in chapter 1.3.

1.2 Insomnia

Most individuals experience a sleepless night at some point in their lives. However, where there is continued re-occurrence and subsequent difficulties in daytime functioning the problem is labelled as chronic. The chapter is concerned with the diagnostic classifications, prevalence, consequences and treatments of chronic insomnia.

1.2.1 Diagnostic Classifications

Two distinct nosologies are currently utilised for the diagnosis of insomnia: Diagnostic and Statistical Manual of Mental Disorders, DSM-IV-TR(86) and International Classification of Sleep Disorders, ICSD-2(2) respectively). Despite subtle differences in the classification of the condition, these two nosologies describe ‘primary insomnia’(86) or ‘general insomnia disorder’(2) as a condition characterised by difficulty initiating and/or maintaining sleep, waking too early and/or non-restorative sleep. Importantly, the complaint is associated with significant distress or daytime dysfunction voiced by the individual. The nosologies diverge at the point where the ICSD-2 further defines this dysfunction, specifying nine possible areas (see Table 2). It further indicates the sleep disturbance must occur despite adequate opportunity for sleep.

Table 2: Description of DSM-IV-TR and ICSD-2 Classifications of Insomnia

DSM-IV : Primary Insomnia (verbatim from 86)	ICSD: General Insomnia Disorder (2)
(A) The predominant compliant is difficulty initiating or maintaining sleep, or nonrestorative sleep, for at least one month	*(A) A complaint of difficulty initiating sleep, difficulty maintaining sleep, or waking up too early, or sleep that is chronically nonrestorative or poor in quality*
(B) The sleep disturbance (or associated daytime fatigue) causes clinically significant distress or impairment in social, occupational, or other important areas of functioning	*(B) At least one of the following forms of daytime impairment related to the nighttime sleep difficulty is reported by the patient:* *a. Fatigue or malaise* *b. Attention, concentration or memory impairment* *c. Social or vocational dysfunction or poor school performance* *d. Mood disturbance or irritability* *e. Daytime sleepiness* *f. Motivation, energy, or initiative reduction* *g. Proneness for errors or accidents at work or while driving* *h. Tension, headaches, or gastrointestinal symptoms in reponse to sleep loss* *i. Concerns or worries about sleep*
(C) The sleep disturbance does not occur exclusively during the course of Narcolepsy, Breathing-Related Sleep Disorder, Circadian Rhythm Sleep Disorder, or a Parasomnia	*(C) The above sleep difficulty occurs despite adequate opportunity and circumstances for sleep*
(D) The disturbance does not occur exclusively during the course of another mental disorder (e.g., Major Depressive Disorder, Generalized Anxiety Disorder, a delirium)	
(E) The disturbance is not due to the direct physiological effects of a substance (e.g. a drug of abuse, a medication) or a general medical condition.	

Contrarily, the DSM-IV makes reference to the duration and the occurrence of the insomnia: for at least 1 month and not exclusively in the context of other sleep disorders, mental and physical conditions or substance abuse. These two additional criteria do however feature in the ICSD-2's description of Psychophysiological Insomnia (PPI), and Paradoxical Insomnia (PI), two of the insomnia subtypes. In addition to meeting criteria for insomnia disorder, PPI is a sleep disturbance resulting from learned sleep-preventing associations and/or heightened arousal. Consequently, the individual will present with one or more of the following: increased sleep anxiety; difficulty initiating sleep at desired times (i.e. no problems when not intending to sleep); improved sleep when away from home and cognitive or somatic arousal in bed.(2)

Although it is one of the most frequently occurring subtypes of general insomnia,(87) the diagnostic category of PPI is not considered particularly reliable or valid, along with the paradoxical subtype(88). Paradoxical insomnia is characterised by a chronic subjective report of little or no sleep that is considerably below age appropriate standards/ does not reveal any homeostatic patterns (rebound nights) and/or in misalignment with objective reports

obtained by polysomnography or actigraphy. The individuals will also describe feeling awake all night with reference to external events and/or excessive thoughts or rumination throughout most of the night. Furthermore the daytime impairment is in discord with the level of sleep deprivation reported.(2)

Idiopathic insomnia on the other hand is considered a more reliable and valid diagnostic entity.(88) Meeting criteria for insomnia and not better explained by another sleep, medical or psychological condition or medication use, the disorder's onset is during infancy or childhood, has no identifiable cause/precipitant and occurs without periods of remission.(2)

Table 3 outlines additional subtypes detailed in the diagnostic nosologies, a comprehensive description of these will not be undertaken in this thesis.

Table 3: Insomnia Subtypes according to DSM-IV-TR and ICSD-2

DSM-IV-TR : Insomnia Subtypes (verbatim from 86)	**ICSD-2: Insomnia Disorder Subtypes** (verbatim from 2)
Primary Insomnia	*Adjustment Insomnia*
Secondary Insomnia due/related to a …Breathing-related sleep disorder	*Psychophysiological Insomnia*
…Circadian rhythm sleep disorder	*Paradoxical Insomnia*
…Dyssomnia Not otherwise specified	*Idiopathic Insomnia*
… a mental disorder	*Insomnia Due to Mental Disorder*
…;a medical condition	*Inadequate Sleep Hygiene*
…Alcohol-related sleep disorder	*Behavioral Insomnia of Childhood*
… a substance	*Insomnia due to Drug or Substance*
…Other sleep disorder	*Insomnia due to Medical Condition*
	Insomnia Not Due to Substance or Known Physiological Condition, Unspecified (Non-organic Insomnia, NOS)
	Physiological (Organic) Insomnia, Unspecified

Because of the limited evidence for some diagnostic criteria, only moderate overlap between nosology classifications,(88, 89) and the majority of cases presenting in concord with another medical or psychiatric disorder(87), there has been a recent development towards eliminating the distinction between 'Primary' and 'Secondary' insomnia. This is replaced with the term 'Insomnia Disorder', whilst simply denoting any comorbid conditions.(90) This change in the fifth edition of the DSM, which is to be published in 2013, is particularly pertinent considering it is often difficult to infer causal relationships in the clinical setting, and brings hope for an eventual convergence of diagnostic nosologies with the eventual publication of the ICSD-3.(91) In 2004 a workforce from the AASM devised research diagnostic criteria aimed to standardise these criteria for insomnia with the hope that these will find their way into the diagnostic manuals.(92) These are outlined in Appendix 1.

This transition also moves away from the implicit assumption in the non-sleep field that insomnia is secondary to other conditions, rather than a disorder in its own right. There is ample evidence to indicate that insomnia can be a predictor of psychiatric conditions, particularly depression(93) following the first longitudinal report in 1989.(94) Furthermore, the treatment of insomnia concomitant to the treatment of depression is associated with larger improvements in depressive symptoms,(95) insomnia may persist long after completion of treatment of only the primary disorder,(96) and individuals with sleep difficulties have poorer outcome trajectories for their depression post treatment, compared to those without,(97) and higher rates of remission.(98)

The largest evidence comes from studies of mood disorders, however there is emerging evidence for similar patterns in other psychiatric conditions.(99)

These concerns of course merit the refinement of diagnostic tools for both the classification of insomnia and differential diagnosis of other sleep disorders. Currently there are a variety of well-validated techniques; these will only be briefly mentioned here, and discussed in more detail (where relevant) in the subsequent chapters. The diagnosis of insomnia is mostly reliant on the clinical interview, for which there are a variety of structured validated tools (e.g. the Structured Interview for Sleep Disorders(100); Duke Structured Interview for Sleep Disorders(101) as well as an insomnia-specific interview schedule.(102) PSG as the gold standard for the assessment of sleep is useful in the differential diagnosis of sleep related breathing or periodic limb movement disorders, but is not considered essential for the classification of insomnia. Less cost and time intensive methods such as actigraphy and sleep diaries are most often used in this population and highlight certain sleep habits or provide information about sleep/wake patterns over longer time periods, particularly useful for the diagnosis of disorders of the circadian rhythm.(103) Both techniques have been validated in insomnia populations against PSG, however actigraphy records tends to underestimate sleep onset latency, and individuals generally misperceive sleep for wakefulness in sleep logs, thus overestimating sleep onset latency and underestimating total sleep time, compared to more objective measures.(104) Self-reported questionnaires, of which the Insomnia Severity Index (ISI,(102)) and Pittsburgh Sleep Quality Index (PSQI(105))are most frequently used, help establish the severity of the insomnia disorder or are used as screening tools.(91)

1.2.2 Prevalence and Consequences

Without consensus surrounding the diagnostic criteria it is perhaps not surprising that there are large variations in the cited prevalence rates of these types of sleep difficulties, ranging from around 6-30%.(87, 106, 107) When only considering a chronic sleep difficulty, with a concurrent complaint of sleep dissatisfaction and daytime impairment and not occurring exclusively in the context of another medical or psychiatric condition or other sleep disorder, the rates tend to be lower. In a recent study, Ohayon and colleagues collected data in seven different countries (France, the United Kingdom, Germany, Italy, Portugal, Spain and Finland) using a validated interview system via the

telephone and concluded that 35% of the sample reported symptoms (difficulty initiating or maintaining sleep, early morning awakenings or un-refreshing sleep ≥3 days a week) 10% reported that these were associated with daytime impairment and 7% met for DSM-IV criteria for insomnia disorder. Primary insomnia (DSM-IV) and psychophysiological insomnia (ICSD-2) were present in 3 and 1.4% of the sample respectively.(87)

A recent epidemiological study in Australia (apart from the UK, the other population this thesis draws samples from) reported similar rates.(108)

The prevalence rates of insomnia tend to increase in older adults to around 20%, again rates depending on the criteria used.(109, 110) Using more stringent criteria this upward trend tends to dissipate, and studies in later older life report lower prevalence rates.(111) Insomnia is generally more common in females than males; a meta-analysis reported a risk ratio of 1.6 for females compared to males and the increased risk was present irrespective of differences in the criteria used.(112)

The consequences of insomnia are numerous, most frequently cited problems are the reduction in quality of life and productivity, and increased risk of accidents and absenteeism.(106, 113) Insomnia may also be a risk factor for various psychiatric conditions as outlined above. Recent studies have also indicated risks of various health problems. Although only cross-sectional, one study showed that individuals with insomnia and objective short sleep ≤ 5 hrs were 500% more likely to have hypertension compared to good sleepers, even after controlling for age, gender, sleep disordered breathing and other potential mediating factors(114). The same working group showed similar results for type-2 diabetes(115) and even mortality in men after controlling for hypertension and diabetes.(116)

1.2.3 Pathophysiology

It is beyond the remit of this thesis to provide a detailed account of the aetiological models of insomnia- this can be found elsewhere.(117) Instead, where relevant to the respective treatments, these models will be briefly introduced in the next section.

1.2.4 Treatment

In becomes evident that insomnia is a debilitating disorder that warrants treatment. To date, the approaches range from complementary/'over the counter' medications to pharmacological, psychological and 'third-wave' therapies and will be outlined in the subsequent parts of this chapter

1.2.4.1 'Over the counter' and prescription medication

'Over the counter' (e.g. antihistamines), alternative treatments (e.g., valerian) and prescription medications (e.g. benzodiazepines, melatonin [in the UK]) are options many individuals with insomnia pursue(107), and these feature most frequently in primary care.(118, 119) However, patients usually indicate a preference for non-pharmacological options(120) and the reason for this paradox is this is largely thought to occur because non-pharmacological options are not readily available.(121)

Prescription medication can be largely divided into those that are specifically indicated for insomnia (benzodiazepine receptor agonists, BZRAs, melatonin [for older adults>55yrs]) by the Food and Drug Administration (FDA) and those approved for other disorders, but used as 'off label' medication for insomnia (e.g. sedating antidepressants).(122) A recent comprehensive review of the literature on short and long-term effects and safety of BZRAs revealed these hypnotics might be beneficial and safe in the short term, however might confer greater risks than benefits in the elderly.(123) They also concluded at the time that limited evidence to determine the long-term safety and efficacy was available. These results echoed the statements issued by both the National Institute for Clinical Excellence (NICE) in 2004(124) and the National Institute of Health (NIH) in 2005,(125) the latter concluding there is little evidence for the use of 'off label' medication and complementary/alternative treatments in this population. Since then the use of BZRAs has increased with off-label medication decreasing.(122)

Pharmacological treatment is largely based on the physiological model of insomnia, which describes elevated autonomic (heart rate, body temperature, heart rate variability, whole body metabolism, skin conductance response) and cortical arousal (increase in higher electroencephalographic [EEG] frequencies; increase in cortical responsiveness/sensitivity to auditory stimuli [event-related potentials]; and poorer 'sleep quality' as determined by cyclic alternating

patterns) as well as increased neuroendocrine (cortisol) output and changes in the neuroimmune response, both during sleep and the day in individuals with insomnia compared to controls.(126)

In conclusion, there is sufficient evidence for pharmacological options in the short, but not long-term. Considering the NICE guidelines indicated the use of hypnotics should occur in the short term only *'after due consideration of the use of nonpharmacological measures'*,(124, p.4) it is necessary to further consider these options.

1.2.4.2 Psychological/behavioural

1.2.4.2.1 Sleep Hygiene

This behavioural strategy is based on the assumption that individuals engage in activities during the day or before bedtime that are sleep incompatible, for example consuming caffeine, nicotine, heavy meals or alcohol and/or exercising too close to the desired bedtime and napping during the day. Unstable sleep/wake patterns are also considered poor sleep hygiene along with a sleep-inhibiting bedroom environment (noise, light, temperature). There is currently no evidence to suggest that the removal of these factors (sleep hygiene treatment) produces meaningful effects and is not recommended as a standalone therapy by the AASM.(103)

1.2.4.2.2 Relaxation Therapy

A number of different relaxation strategies have been developed to deal with the somatic hyperarousal characteristic of insomnia described above. These include progressive muscle relaxation,(127-129) passive relaxation (diaphragmatic breathing, body focusing, autogenic phrases),(130) and biofeedback.(131, 132) Progressive muscle relaxation is probably the most frequently referenced technique; it includes the progressive tensing and relaxing of each major muscle group. Passive relaxation is similar to the above, with focus on full body relaxation, but excluding the element of tension.(133) Biofeedback encourages relaxation through amplifying physiological arousal through visual or auditory stimuli; in the earlier studies individuals were trained to reduce the tone of the auditory feedback linked to their level of relaxation.

Imagery is often considered a type of relaxation, however this, contrary to biofeedback and progressive muscle and passive relaxation strategies, are not provided guideline or standard level of approval, respectively, by the AASM practice parameters.(103)

Some of these techniques are not confined to the reduction of somatic arousal, but may also target cognitive arousal featured in other models of insomnia outlined below.(102, 134-136)

1.2.4.2.3 Sleep Restriction Therapy

In 1987 Spielman described a technique designed to deal with one of the perpetuating behaviours individuals with insomnia engage in: extending time in bed.(137) In an effort to compensate for prior wake-filled nights, individuals tend to go to bed earlier, stay in bed later and possibly nap during the day. These maladaptive behaviours, can perpetuate the initial sleep complaint, pushing someone over the threshold from acute to chronic insomnia, according to Spielman's three-factor model.(138) These perpetuating factors (behavioural component) contribute to the development of a chronic problem in addition to the predisposing (genetics, personality) and precipitating (stressful live events) variables (stress-diathesis component). Sleep restriction, as described by Spielman, is designed to break the perpetuating cycle by reducing the time individuals spend in bed. The prescribed time is modelled on the individual's average sleep time obtained from the sleep diary. The sleep restriction window is reduced by setting a morning time in concordance with the patients circadian and social/occupational preferences and subtracting the given average total sleep time, to establish the threshold time, after which the individual may retire when feeling sleepy. This sleep window is maintained for at least 1 week and encourages re-establishing the homeostasis of the sleep wake-process. The reduction of time in bed produces minor sleep deprivation and thereby 'jump starts' the homeostatic drive for sleep. A constant routine also helps regulate the circadian sleep-wake cycle. At subsequent sessions the window is titrated on the basis of improved and consolidated sleep. The window is extended in steps of 15-30 minutes once sleep efficiency (the percentage of time in bed spent asleep) reaches 90% or more. As most individuals will present with a low average total sleep time (approx. 5-6 ½ hours), many will struggle to follow this

routine.(139) Nevertheless, when this treatment is implemented in isolation, it can produce meaningful improvements that are comparable to other behavioural techniques.(140) However, at the point of publication of the AASM practice parameters, there was only sufficient evidence to endorse this treatment component as guideline, but not as a standard treatment.(103)

A variant of sleep restriction has been explored by Lichstein and colleagues, termed sleep compression.(130) Rather than immediate restriction, this technique invovles the gradual reduction of time in bed over the course of the program, ultimately reaching the baseline total sleep time. Weekly reductions are calculated based on the difference between baseline sleep time and time in bed, divided by the number of treatment weeks. Titration based on sleep efficiency is made akin to sleep restriction therapy. Sleep compression is compatible with Lichstein's biodevelopmental model, describing insomnia as a discord between developmental changes and sleep expectation,(130) thus there is uncertainty whether this would be effective for individuals with paradoxical insomnia or where the reduced sleep time is not a result of developmental changes.

1.2.4.2.4 Stimulus control

This therapy, along with relaxation strategies and cognitive behaviour therapy is the only treatment to have been evaluated as standard for the treatment of chronic insomnia by practice parameters.(103) Although classic conditioning principles may contribute to the development of chronic insomnia,(117) this treatment is based on the notion that the sleep problem emerges because of poor stimulus control (operant conditioning). Adapted from principles of behaviourism, Bootzin describes the sleep environment of the insomnia patient as characteristic of stimulus dyscontrol.(Bootzin, 1972 as cited in 117) During good stimulus control, the desired response (sleep onset) is evoked by the exclusive pairing of the bedroom environment (bed, bedroom, bedtime) as a discriminative stimulus and sleep (reinforcer). In contrast, during stimulus dyscontrol, the bedroom environment is no longer discriminative for the occurrence of the reinforcement. This may be because of a lack of strong discriminative stimuli, or the presence of sleep prohibiting stimuli (watching TV, reading, working). The goal of this therapy is to eliminate sleep incompatible stimuli from the bedroom environment (e.g. no TV/reading/working in bed) and

re-establish strong discriminative stimuli for sleep (e.g. do not sleep/nap outside of bed) i.e. establishing good stimulus control. The addition of the quarter-of-an-hour rule, instructing individuals to get up out of bed when awake for more than about 15 minutes, removes the wakeful/worry/rumination aspect from the sleep environment in addition to the other wakefulness promoting behaviour.

The efficacy of stimulus control was recognised in the earliest meta-analysis, with effect sizes for sleep onset latency (SOL), wake after sleep onset (WASO), and number of awakenings at night (NWAK) of d=0.8; 0.7 and 0.59 respectively.(140) Stimulus control appears as a component of most cognitive behaviour therapy packages and also features, alongside sleep restriction, in the brief versions.(118, 141)

1.2.4.2.5 Cognitive Therapy

This approach in general aims to target and replace misconceptions, maladaptive attitudes towards and beliefs about sleep as outlined in the models of insomnia explicitly describing cognitive factors contributing to the aetiology and maintenance of the disorder.(102, 135, 136) Initial conceptualisation of the cognitive model of insomnia was provided by Morin in 1993(102) and extended by Harvey in 2002.(135) According to Morin dysfunctional cognitions are an integral part of the intricate, self-perpetuating relationship between arousal (cognitive, physiological and emotional) maladaptive behaviour (e.g., napping), consequences (e.g., low mood, fatigue) and the disorder. Thus, targeting these beliefs and attitudes is an essential addition to the behavioural strategies; cognitive therapy includes the following steps:

-Outlining the link between thoughts and emotions

-Identifying and challenging maladaptive attitudes and beliefs

-Working with the patient in replacing these with more adaptive thoughts.

Strategies and techniques used to accomplish these are borrowed from cognitive therapy as outlined by Aaron Beck and include for example Socratic questioning or guided discovery.(142) Harvey describes how these maladaptive cognitions in combination selective attention, safety behaviour and misperception of sleep loss and daytime effects can lead to a real deficit during night *and* during the day and thereby maintain the disorder. This model thus

gives equal weighting to night and daytime effects, in line with the view that insomnia is a 24hr disorder.(135)

Additional steps to cognitive therapy thus also include targeting sleep misperception, eliminating safety behaviour like napping, and reducing attention bias, focusing explicitly on the daytime affects in addition to nighttime.(143) It might also include techniques, such as thought management (e.g. imagery(144) or simple strategies like saying the word "the" to occupy mental capacity).

In comparison to the behavioural techniques outlined above, the literature on the efficacy of cognitive therapy for insomnia is less extensive. There is currently no randomised trial comparing the efficacy of cognitive therapy versus behavioural techniques. However a clinical trial is currently ongoing comparing cognitive therapy (cognitive restructuring) with behaviour therapy (SRT and SC) and CBT-I,(145), its efficacy has been documented in an open trial,(143) and there is evidence that maladaptive beliefs are sensitive to change with CBT-I.(146-148)

1.2.4.2.6 Paradoxical Intention

Initially adapted for sleep-onset insomnia(149, 150) from the original work by Frankl(151) this technique was aimed at targeting the patients increased effort to sleep and related anxiety when unable to "perform".(152) Patients are instructed to lie in bed, keep their eyes open for as long as possible and give up the active effort for sleep. Espie adds a cognitive component to paradoxical intention instructing the individual to accept wakefulness, seeing it as an opportunity rather than tragedy.(152) This treatment is in concord with Espie's psychobiological inhibition(153) and Attention-Intention-Effort(136) models, which consider sleep normalcy as automatic and involuntary and insomnia as a disruption to that process. In the A-I-E model, Espie further refines the components that contribute to the inhibition of this normal process: a) the selective attention towards b) explicit intention to and c) increased effort to initiate sleep.(136) In addition to paradoxical intention, both models sit well with other therapies described above (e.g. cognitive therapy and stimulus control).

1.2.4.3 Cognitive-Behavioural Therapy for insomnia (CBT-I)

Cognitive behavioural therapy for insomnia (CBT-I) is a multi-component therapy that combines single or multiple behavioural and cognitive strategies outlined above. A total number of 8 meta-analyses have summarised the effects of CBT-I for improving sleep related outcomes.(140, 154-159) These meta-analysis indicated medium to large effect sizes (Cohen's d(160)) at the end of treatment for SOL [0.4-1.0], WASO [0.6-1.0], sleep efficiency (SE) [0.5-0.8], NWAK[0.25-0.8], and medium for total sleep time (TST) [0.2-0.5] and large effect sizes for sleep quality (SQ)[0.7-1.5]. The magnitude of change tends to increase at follow-up, especially total sleep time when studies include a sleep restriction component.

The only model that explicitly combines the psychological and biological perspectives is Perlis' neurocognitive model(134) and is worth mentioning at this point. In an attempt to explain phenomena such as sleep misperception, Perlis describes the development of insomnia as a result of conditioned cortical arousal. The initial precipitating event might induce cognitive, somatic and cortical arousal as a result of the stressor. In line with the behavioural perspective, chronic insomnia develops and is maintained through maladaptive behaviours like extending the time in bed; as a result, the bedroom becomes associated with arousal through classic conditioning principles, absent from the original stressor. The increased cortical arousal at sleep onset explains changes in sensory and information processing as well as long-term memory formation and contributes to the individual's (mis)perception of increased wakefulness and decreased sleep. This phenomenon is characteristic of individuals with insomnia and to an exaggerated extent in paradoxical insomnia.(161) Thus targeting cognitive, behavioural and physiological aspects of the development of insomnia with CBT-I fits well with this model.

1.2.4.4 'Third-wave' techniques

Interesting 'third-wave' techniques have been developed in addition to the CBT-I techniques and pharmacological options mentioned above. Mindfulness-based stress reduction is an approach that encourages the individual to adopt an accepting and non-striving approach to sleep and the negative thoughts they might endorse, subsequently reducing the somatic and cognitive arousal and its

efficacy has recently been tested in combination with cognitive-behavioural techniques.(162)

Another exciting development is intensive sleep retraining (ISR) developed by a group in Adelaide, Australia.(163, 164) It is based on classic conditioning and involves retraining the already sleep deprived (5hrs on pre-treatment night at home) individual by repeated waking after 3 consecutive minutes sleep. This is continued throughout all 50 sleep opportunities over a 25 hr period. A recent randomised control trial comparing ISR with or without stimulus control to stimulus control alone or control (sleep hygiene) reported despite all active arms producing significant improvements, the arms including ISR had speedier improvements in SOL and TST by week one, compared to the arms without.(164) Lastly, a technique presented at the Associated Professional Sleep Societies (APSS) conference in 2011 involved cooling of the frontal cortical areas, which have been associated with increased activity in the individual with insomnia.(165) This was only a small study of 12 participants with primary insomnia, matched with 12 healthy controls, thus larger studies are warranted.

One novel development worth mentioning is the combination of pharmaco- and psychological therapies, which might complement each other, and utilise their respective strengths (fast acting vs. sustained effects respectively).(166) A recent study tested the delivery of CBT-I alone or in combination for 6 weeks, which was further extended by continued CBT-I alone, no treatment or no change to treatment (i.e. continued zolpidem+CBT-I). The best treatment response was for the combined arm followed by CBT-I alone.(167) Further research in these areas is clearly warranted.

1.2.5 Summary

It is evident that chronic insomnia is a prevalent and debilitating disorder that warrants treatment, either with pharmacological (short-term or in combination with CBT-I), cognitive and/or behavioural treatments. The efficacy has been established, but to determine true effectiveness, it is now important to turn the attention towards adherence.

1.3 Adherence

"Keep a watch also on the faults of the patients, which often make them lie about the taking of things prescribed" Hippocrates (460–377 BC)

1.3.1 Definition of Adherence:

As described so potently by Hippocrates himself, adherence is a natural human process that yields considerable attention. Especially because adherence to medical recommendations is one of the strongest predictors of treatment outcome health practitioners need to consider managing and promoting adherence as part of treatment itself. The World Health Organisation (WHO) recently emphasised that increasing adherence to existing treatments can have a far greater impact on health than improvements in specific treatments for disorders.(168)

As reported in a recent meta-analysis, the average non-adherence rate of approx. 25% for complex treatment interventions translates to 188.3 million medical visits in the US resulting in patients failing to implement recommendations and equating to a monetary waste of potentially US$ 300 billion a year.(169) Apart from this economic cost, non-adherence inhibits the evaluation of treatment efficacy at the research level, and can hinder treatment effectiveness at the clinical level.

Reflective of this impact, there has been an increase in research on adherence to medical regimes and recommendations in the last decades. In this journey to understand complex issue of adherence, a large variance in reported adherence rates has been cited. These differences may be accounted for by illness and treatment type and the assessment method of adherence.(169) A further reason, only briefly touched upon by the author of the meta-analysis, may be differences in conceptualisation of the term adherence. Although the terms adherence and compliance have often been used interchangeably in research, there are distinct differences that have been recognised. Compliance refers to *"the extent to which a person's behaviour (in terms of taking medications, following diets or executing lifestyle changes) coincides with medical or health*

advice",(170, pp. 2-3) whereas the use of the term adherence implies that the individual's behavior "*corresponds with agreed recommendations from a health care provider*"(168, p.18)

Thus the transition away from the term compliance emphasises the importance of a non-authoritarian approach and a balanced doctor-patient relationship.(171) In order to accomplish this in a clinical setting, the practitioner is required to gain insight into the patient's beliefs and attitudes towards changing their behaviour, and to understand which factors play a role in motivation to adhere. At the research level, psychosocial variables are considered over and above the static biomedical factors, see Table 4.

Table 4: Conceptualisation of adherence vs. compliance

Level	Compliance	Adherence
Conceptual	Person's behaviour *coincides* with medical or health advice given	Person's behaviour is in correspondence with the *agreed* recommendations
Research	Biomedical (Biological/treatment specific/demographic) variables	Psychosocial variables (beliefs, attitudes, motivation, social support) and biomedical factors
Clinical	Biomedical model	Patient-centred/ Holistic/ Integrative model (BPS)

Some have argued the term adherence still implies a sense of authority,(171) and there has been push towards the term 'concordance', which is more focused on the success of involving the patient in the decision making process.(172) The term adherence will be used in the remainder of this thesis because it is frequently cited in the literature, and sufficiently differentiates itself from the medical approach to the patient's behaviour. The exception to this is in chapter 5, which has been included as a published paper, where the term compliance is used to describe the numeric value rather than the behaviour). This introductory chapter will review the literature on adherence to CPAP and CBT-I. Three areas will be covered for each treatment: 1) measurement and 2) predictors of and 3) interventions to improve adherence, with a brief introduction to medication adherence at each section, because this is undoubtedly the most frequently researched topic.(169)

1.3.2 Measurements of Adherence

Measurement of adherence to medication can largely be separated into two categories: indirect (self or family/physician report, markers such as pill counts or filled prescriptions and electronic monitors) and direct (urine/blood tests).(173). Electronic monitors and urine/blood tests are often considered gold standard,(174) however these are costly and time-consuming; reported estimation of adherence is often above objective measurements when reported by the practitioner(175) and patient.(176) Due to the large variety in types of measurements, average rates of non-adherence are difficult to obtain, however a recent meta-analysis reported an adherence rate of 79.4%.(169)

1.3.2.1 Measurement of Adherence to CPAP

1.3.2.1.1 Subjective Adherence

The most common metric for CPAP adherence reported in the literature has been 'average hours of CPAP use per night'. There are however some variations, which include 'percentage of days CPAP was used', 'percentage of study participants considered adherent using the cut-off of at least 4hrs CPAP use/night for at least 70% of nights/week'; 'percentage of nights used more than 4hrs' or 'average CPAP use per night only considering nights used'. (177) Throughout this thesis, where not otherwise specified, adherence to CPAP refers to the common metric of 'average CPAP use in hours/night'.

The earliest empirical studies assessing CPAP adherence rates relied exclusively on subjective self-reported use. The initial consensus was that subjective adherence was relatively high and CPAP was a well tolerated therapy for the treatment of OSA,(178-180) however subsequent reports found these subjective rates to only weakly correlate with objective adherence and reported that individuals overestimate actual average nightly CPAP use by as much as 1 hour.(181-183) In Kribbs seminal prospective study in 1993, thirty-five individuals with OSA were provided with a CPAP machine and followed up an average of 3 months later.(181) A microprocessor that established mask-on-face time, indicated that individuals used the machine on average 4.88 +/- 1.97 hrs/night. However, when asked to report their use subjectively, this rate was increased by 69 +/- 110 min. These differences were replicated in Engleman et

al. 1996(182) and Rauscher et al. 1993,(183) who reported differences between 0.9 hrs and 1.2 hrs/night respectively.

1.3.2.1.2 Objective Adherence

Since the initial reports on these covert electronic surveillance in 1993 and 94(181, 184), recent studies have almost exclusively relied on in-built time counters assessing 'objective' or microprocessors measuring 'effective' use, indicating that CPAP adherence is not as high as initially thought. Earliest studies evaluating CPAP use other than through self-report, used inbuilt time counters that recorded the time the machine was turned on for.(185, 186) However, this technique was not considered optimal, with the fear that individuals might be aware of being monitored(181) Additionally, it allowed no measure of the time the mask was effectively worn, and it was not until the simultaneous publication of the use of a technique to measure pressure at the mask from two separate working groups,(181, 184) that this was accomplished. A pressure transducer located on the air outflow of the machine records the rise in pressure when the mask on the face thereby recording effective CPAP use.(181) Although these two measurements (objective use and effective use) are strongly correlated(187) with only a 10 % difference in these recordings(181), the majority of studies today report effective use obtained from a smart card located within the CPAP device.

With these methods of assessment, it is generally accepted that adherence to CPAP is less than adequate. Starting with initial acceptance, studies have indicated that approximately 1/4 of patients do not take up CPAP or discontinue in the first 2 weeks after their titration study.(188-192) Rauscher et al. reported that of 95 individuals with OSA, who had been offered CPAP, 32 % rejected CPAP immediately and of those who did 27% refused home treatment shortly after the titration night. Similar post titration acceptance rates between 20-30 % were reported in a subsequent review(190) and again replicated in more recent studies.(188, 189, 192)

The majority of individuals who use CPAP at 1 month will continue doing so long term and early use is a strong predictor of long-term adherence (see below). However objective studies of those continuing treatment (>1month), 'adequate' adherence rates (often defined as ≥ 4hrs on 70% of the nights(181)) can vary between 46-89% depending on definitions of adherence and length of

follow-up.(181, 184, 188, 193-202) The average hours of use per night in these studies varies between 4 and 6 hrs. Most of the above studies have used the arbitrarily set cut-off of 4 hrs per night on 70 % of the nights for 'adequate' adherence based on Kribb et al's work in 1993,(181) and thus adherence is often conceptualised as a dichotomous variable (adherent vs. non-adherent). This process was initially validated with two distinct patterns emerging from analysis of CPAP users,(203, 204) however recent evidence challenges this approach. Aloia et al. examined CPAP use in 71 OSA participants across 365 days and reported a total of seven categories of adherence, including the compliance/non-compliant group, but adding slow improvers/decliners, variable users, occasional attempters and early drop-outs.(205)

In addition, recent work by Weaver et al. in 2007 questions the arbitrary cut-off of 4hrs/night. The authors highlighted different values for optimisation depending on outcome variable. The majority of individuals returned to normal values for daytime sleepiness after an average 4hrs/night of CPAP use, whereas for objective sleepiness and daytime functioning, the values were 6 and 7.6 hrs/night respectively. There was a dose-response relationship between CPAP use and improvements in sleepiness, indicating more use leads to a better outcome of increased alertness.(57)

1.3.2.2 Measurement of Adherence to CBT-I:

The information on measurement of adherence to CBT-I is much less abundant. This may to a large extent be due to its multicomponent nature(206) and the majority of studies have focused on examining adherence to the behavioural components of CBT-I, whether these are presented in isolation or within the cognitive-behavioural package (see Table 5). Furthermore, CBT-I may inherently incorporate efforts to monitor and improve adherence.(see 207)

Measurements are largely restricted to very crude measures such as attendance rates or self/spouse or therapist questionnaires, some of which are poorly defined or designed.(e.g. 208, 209-211) One study(208) adapted the five-item Medical Outcomes Study Specific Adherence Scale(212) to assess adherence to sleep restriction and stimulus control. Three of the five items assessed individuals' adherence to the treatment plan (e.g. I was unable to do what was necessary...") however two of the items assessed 'ease' and 'difficulty' in

adhering to the doctors recommendations, aspects that are not exclusively associated with adherence. This might explain the moderate Chronbach's alpha of .78 reported by the original authors.

A more reliable measure perhaps has been the use of sleep diaries to obtain an estimation of deviation from their prescribed threshold and rising time and time in bed. In 2001 Riedel and Lichstein published a detailed description of 5 strategies to calculate adherence to a sleep compression program.(213) No changes were made to the bed window at the final treatment session, so adherence was could also be calculated from the 2-weeks posttreatment sleep diary. The following 5 adherence measures were computed: (1) difference from prescribed time in bed (TIB) at final treatment to reported TIB at posttreatment. (2) reported TIB reduction as a percentage of prescribed TIB reduction (3) mean TIB change (4) consistency of TIB (5) consistency of get up time. This study found a relationship only between the latter two measures and treatment outcome (fewer and shorter awakenings and sleep quality improvements), indicating consistency rather than absolute minute adherence was related to outcome. Other studies have described a positive relationship between various outcome measures and therapist rated(214) or self-reported/diary obtained adherence,(137, 206, 208, 215-217) however others have found no relationship(218) or only non-significant trends.(219)

Apart from one study examining the optimal "dose" of CBT-I sessions,(220) there has been no dose-response study to determine how much of any or each component is needed to produce significant improvements.

There are a few unique issues to consider with CBT-I that are worth mentioning. The first is relative adherence, that is, how adherent was the individual at times when implementation was required. Importantly, with some components (e.g. thought blocking; quarter-of-an-hour rule), increased improvement is associated with a decrease in required implementation. Some studies outlined in Table 5 have not factored this in to their adherence measure, (210, 211, 221-223) in others it remains unclear;(206, 209, 219, 224) overlooking this consideration may result in an overestimation of non-adherence.

Secondly, and this concerns the measures outlined in Riedel and Lichstein(213) and used by others,(162, 208, 214) when assessing adherence to sleep restriction, there is a need to score bed and get up times within the set window as adherent e.g., if an individual is told to go to bed at 1am and retires

at 1.30 am when feeling sleepy, this should not be considered non-adherence. Perlis et al. 2004(218) for example replaces all time points after the threshold time to zero, indicating adherence to the sleep restriction instructions ("go to bed on or after threshold time"). This has not been incorporated in Riedel and Lichstein's calculations, failure to do so for both threshold and rising time (at least when sleep restriction therapy is administered without stimulus control, where the individual is instructed to keep a consistent rising time), could overinflate rates of non-adherence.

Table 5 outlines studies that have examined either single or multiple behavioural components and CBT-I. Definite conclusions cannot be drawn because of the differences in study designs, recruited samples and adherence measures. In summary, a wide range of techniques has been presented to measure adherence to components of CBT-I. Thus, it is difficult to draw any conclusions about optimal adherence from this data. Nevertheless, it does seem that adherence in this population is skewed towards higher rates, and where adherence to sleep scheduling is conceptualised in minute deviation, individuals are mostly within 30 minutes of their prescribed times, see Table 5. Where individual components are compared, adherence rates for sleep hygiene and in some studies relaxation seem to be highest. Undoubtedly, our understanding of adherence to the cognitive and behavioural treatments for insomnia is relatively sparse, especially in terms of 1) optimal measures of adherence 2) possible normative values or clinically meaningful cut-offs for 'adequate adherence' 3) any dose-response relationship with outcome. This information could have significant implications for the evaluation of CBT-I effectiveness, this being a function of treatment efficacy and patient adherence.(168, 225, 226)

Table 5: Overview of studies measuring adherence to CBT-I

	Author (Date)	Sample Description*	Adherence Measure	Predictors of Adherence (Association with adherence ↑↓)	Adherence Rates
Single/Multiple Behavioural Components	Spielman et al. 1987(137)	35 CI completing sleep restriction therapy	Sleep Diary [less time time in bed reduction at follow-up compared to posttreatment]	-	Follow-up reduction in TIB was less (39 minutes) than at posttreatment (74 minutes)
	Schoicket et al. 1988(209)	74 SMI randomised to sleep hygiene (SH), meditation (M) or stimulus control (SC)	Self-report [number of days components were adhered to]	-	SH= 5.5 days per week M= 5.6 SC= 4.7 days
	Riedel et al. 1995(227)	RCT with older PI and older adults without insomnia (WoI); 50 randomised to education video and 50 to video+guidance on sleep compression (video included guidance on restricting time in bed)	Sleep Diaries [significant reduction of time in bed at posttreatment and followup]	-	Education+video= significant reduction of time in bed to posttreatment (PI and WoI) and follow-up (PI only) Video only= no significant reduction of time in bed
	Riedel & Lichstein 2001(213)	22 older PI completing SRT	Sleep Diary [5 measures, variant calculations of minute deviation from prescribed time in bed]	-	Individuals spent 28 min more in bed than prescribed, 64% were in bed within 30 min and 36% within 15 min of their time in bed.
	Pallesen et al. 2003(219)	RCT with 55 older CI randomised to either sleep hygiene+stimulus control or sleep hygiene+relaxation	Self-report [rating from 1-3 on adherence to all components with higher scores indicating greater adherence]	-	-
	Waters et al. 2003(211)	53 CI randomised to either SH, M, SR/SC or progressive muscle relaxation + Cognitive distraction training (PMR/CD). In phase 2 participants in the PMR/CD group were also provided with SR/SC instructions	Self report [Adherence Checklist asking participants if they adhered to the relevant component with a yes/no response format, adherence was conceptualised as the % of nights the recommendation was followed]	-	Participants adhered to the components between 47-99% of the nights. The highest adherence rates were reported for medication and lowest for cognitive distraction.

	Carney et al. 2004(228)	68 good sleepers randsomised to being made aware or not that the actiwatch monitors adherence to SRT	Actigraphy [Bedtime and Get-up time were determined from algorithms to establish a reduction or increase respectively in activity respectively]	-	Aware group: went to bed 12 minutes after their threshold time; got up 16 minutes after Non-aware group: went to bed 1hr12 min late; got up 30 minutes late.
	Epstein et al. 2012(229)	RCT with older PI; 44 randomised to SC, 44 to SRT, 41 multi-compoment treatment (SC+SRT)	Attendance rates [attended out of 6 sessions] Self- & significant other report [ratings on scale of 0-4 at posttreatment and follow-up with higher scores indicating better adherence; Sleep Diaies [self-report questions to obtain % of days components were adhered to]	-	Significantly more session were attended in the SC and multi-component arm= 5.86 and 5.83 respectively, than the SRT arm. Average self- and spouse reported adherence rates were ≥3 in all treatment arms Sleep diary ratings were 89.3% for CBT-I; 87.3% for SRT, and 90.0% for multi-component.
	McChargue et al. 2012(221)	113 breast cancer patients with co-morbid insomnia completing behaviour therapy (SC, SRT, relaxation and sleep hygiene)	Self-report [individuals indicated behavioural treatments used on all days assessed]	Increase in depressive symptoms ↓ Improvements in sleep ↓	Individuals implemented behavioural components 51-52% of nights measured. Highest rates reported for sleep hygiene, lowest for sleep restriction and stimulus control
CBT-I	Mimeault et al. 1999(224)	RCT with 36 PI randomised to bibliotherapy either with or without therapist guidance	Self & Significant Other Report [adherence items on treatment evaluation questionnaire; ratings ranged from 0-100 with higher score indicating better adherence]	-	-
	Vincent et al. 2001(230)	37 CI completing CBT-I	Attrition [Completed or did not complete treatment]	Pre-treatment acceptability ratings of CBT-I treatment↑	Dropouts= 18%
	Harvey et al. 2002(206)	90 PI 12 months after completion of CBT-I	Self-report [% of individuals indicating the continued use of 10 components]	-	On average 39% of individuals were using components; highest reports for relaxation=74%; lowest for imagery=19%
	Bouchard et al. 2003(231)	39 PI	Sleep Diary [Composite Score of adherence to 8 behaviours obtained from the sleep diary entries, scores ranged from 7-49 for each week with higher scores	General, task-related and self-regulatory self-efficacy ↑	Average score over all weeks=44

		indicated better adherence]		
Morgan et al. 2003(232)	RCT with 108 chronic hypnotic users randomised to CBT-I	Attendance Rates [individuals were separated into those completing all and fewer than six sessions]	Poorer sleep quality (PSQI) at baseline ↑	65% completed all 6 therapy sessions
Vincent et al. 2003(214)	50 PI	Sleep Diary [consistency of bedtime and wake-up time] Therapist and Spouse reported [scale 5-25 with higher scores indicating greater adherence] Attendance Rates [% Therapy Sessions Attended]	Dysthymia* ↓	Attendance Rates 92% Therapist reported 48% were very much or extremely adherent Average spouse-report = 21
Perlis et al. 2004(218)	RCT with 10 PPI to Modafinil+CBT-I and 12 PPI to Placebo+CBT-I	Sleep Diary [adherence was considered if the individual went to bed on average 15 minutes earlier than prescribed time for each week; final adherence score=% of individuals adherent each week]	-	CBT-I+modafinil= 80% adherent CBT-I+placebo=51% adherent
Vallieres et al. 2005(233)	RCT with 17 PI randomised to three different combinations of CBT-I and medication	Sleep Diary [see Bouchard et al. 2003; scores were presented in percentages]	-	Sequence 1: CBT-I + Med after medication alone; rates of behavioural components ranged from 68 .3% to 92.5% across weeks Sequence 2: Combined med and CBT-I rates were 92.3% and decreased to 72.5% in subsequent CBT-I arm Sequence 3: CBT-I alone rates varied between from 50.0% to 85.7%.
Sivertsen et al. 2006(210)	RCT with 18 PI randomised to CBT-I	Attendance Rates [number of sessions attended] Self-report [adherence ratings on a scale 0 (never) to 5 (every day)]	-	Attendance rates 100% Average rating= 4.8 (6-week posttreatment); 4.1 (6-mth follow-up)
Ong et al. 2008(162)	30 PPI undergoing CBT-I+mindfulness	Sleep Diary [see Riedel and Lichstein, calculated adherence to time in bed and get up time]	-	22 minutes more time in bed 30 minutes in bed later than get-up time
Vincent et al. 2008(208)	40 CI completing CBT-I	Attendance Rates [%therapy sessions attended]	Increased age*↑ Use of medication*↓	91% sessions attended Average score of 21.4 on self

		Self-Report [Global ratings ranging from 5 to 30] Sleep Diary [consistency of wake-up time]	Perceived certainty in adherence*↑ Fewer barriers ↑ Pretreatment sleepiness ↓	report Variance in wake-up times post treatment= 0.91hrs
Morin et al. 2009(167)	RCT with 38 PI to CBT-I (6weeks)+extended CBT-I (6months); 37 to CBT-I +no treatment; 37 to CBT-I +zolpidem+extended CBT-I alone and 37 to CBT-I +zolpidem+extended CBT-I with zolpidem as needed	Attendance Rates [Therapy sessions]	-	6-weeks CBT-I = 5.6 sessions attended 6-week CBT-I +zolpidem= 5.8 6 month extended CBT-I =5. 5 6 month CBT-I with no further zolpidem=5.4 6 month CBT-I with zolpidem as needed=5.5
Trembley et al. 2009(216)	RCT with 57 cancer patients with co-morbid insomnia randomised to CBT-I	Sleep Diary [dichotomous evaluation (yes/no) of sleep diary entries along 5 criteria; adherence depicted the % of days individuals adhered to these criteria)	-	88% at both 8-week posttreatment and 6 months follow-up
Vincent et al. 2009(222)	RCT with 59 PI randomised to online CBT-I	Self-report [% of individuals implementing components ≥4 nights per week]	-	Lowest adherence rates =hypnotic tapering (22..6%); highest adherence rates=sleep hygiene(76.8%); Of relaxation strategies: Paced breathing > progressive muscle relaxation, hypnosis or imagery
Hebert et al. 2011(223)	47 PI completing online CBT-I	Self-report [see Vincent et al. 2009]	Perceived behavioural control*↑ Social support*↑ Intention to complete CBT-I*↑ Readiness to change*↑	Week 1 range= 39% (exercise)-95% (alcohol tapering) Week 2= 82.6 % (avoid napping, regular sleep schedule, avoid reading in bed) Week 3 range= 39% (muscle relaxation and imagery)-60%(abdominal breathing)
Manber et al. 2011(215)	301 CI completing CBT-I in a clinical setting	Self-report [assessing the degree (0-3) of adherence to all components of CBT-I-higher scores indicate better	Depression scores* ↓	Behavioural Components: mean 2.2 (Low Depression Group) mean= 2.1 (High Depression Group)

		adherence]		Cognitive Components: mean=2.3 Low Dep Group); mean=2.2 High Dep Group)
Matthews et al. 2012(217)	34 breast cancer patients with co-morbid insomnia undergoing CBT-I	Sleep Diary [Daily yes/no evaluation according to 1) bedtime 15 within prescribed time; 2) get up time within 15 minutes of the prescribed time 3) total time in bed within 30 minutes of prescribed time; adherence was defined as days adherent/week]	type of cancer treatment received (Chemotherapy)*↑ motivation to change* ↑ baseline fatigue* ↓	Adherence to 1)bedtime= 5.4-6.1 days/week across all treatment weeks 2) rise time= 3.0-4.1 3) time in bed= 4.2-5.1

CI= Individuals with Chronic Insomnia; PI= Individuals with Primary Insomnia; PSQI= Pittsburgh Sleep Quality Index; PPI= Psychophysiological Insomnia; RCT= randomised Controlled Trial; SMI= sleep maintenance insomnia; * correlational or test of differences only (not regression analysis); $^{+}$ Randomised individuals; overall sample size might be larger, where RCT included a non-CBT-I (e.g. medication) arm or placebo/waitlist control; SC=Stimulus Control; SRT= sleep restriction therapy. Studies were identified through a PubMed search with the terms “adherence” or “compliance” and “insomnia” and “CBT” and through reference lists of other publications.

1.3.3 Predictors of Adherence

Establishing predictors of adherence is important for informing potential interventions to increase adherence. A recent comprehensive review of medication adherence reported certain factors to be associated with better adherence: increased age, positive beliefs about the illness and treatment, social support, positive doctor-patient relationship; whereas other factors are generally barriers for adherence: psychological status (e.g. depression, anxiety, stress, low self-efficacy), some sociodemographic factors (low literacy, no insurance and homelessness), adverse events, low English proficiency/health literacy and regime complexity.(234)

1.3.3.1 Predictors of Adherence to CPAP

1.3.3.1.1 Early CPAP use

CPAP adherence within the first month is a strong predictor of long-term use (1 month-12 months) with the majority of studies reporting very strong Pearson's correlations of .7-.8, where reported.(181, 188, 196, 199, 201, 203, 204, 235-239). Aloia and colleagues showed CPAP adherence at 3 months in a sample of 140 OSA patients could be predicted by machine use on the first day. The Pearson correlation between CPAP adherence at day 1 and 6 months was moderate (r=.35), 61 % of >4hr users on day 1 were also considered adherent at 6 months.(204)

Other factors can be separated into biomedical, psychological and social factors as outlined in Table 6 along with references.

1.3.3.1.2 Biomedical Factors

1.3.3.1.2.1 Patient-Specific

1.3.3.1.2.1.1 Age/Gender/Race

Most studies indicate weak if at all clinically relevant relationships between age and CPAP adherence, especially when other factors are controlled for in multivariate analysis (e.g. gender, baseline ESS, baseline AHI), the relationship between age and

adherence disappears.(240, 241) A recent review however identified factors that might be unique to paediatric and geriatric groups.(242) A similar effect has been shown for gender, with some studies indicating higher rates in females, others in males and others again indicate no difference. Often these differences are too small to be considered clinically relevant; for example, a difference of 7 minutes/ night was reported between males and females in one study.(243) Limited information is currently available to make reliable conclusions on the effects of race or ethnic group. The majority of these studies have compared African American with Caucasian race and most have found higher adherence rates for Caucasians. However, some of these effects observed may be mediated by socio-economic status, as found in one study comparing lowered adherence rates in 25 Maori to the increased use in 101 non-Maori CPAP users.(244)

1.3.3.1.2.2 Treatment-Specific

1.3.3.1.2.2.1 Pressure modification

Although the level of pressure (cm H_2O) itself does not seem to have any impact on CPAP adherence, a number of studies have investigated the effect of different pressure delivery modes (i.e. autoPAP, BiPAP, and flexible pressure delivery (C-Flex)). Three recent meta-analyses(177, 245, 246) of technological interventions investigated pressure delivery modifications (i.e. autoPAP, BiPAP, and flexible pressure delivery (C-Flex)) yet found no evidence for increased adherence in any modality except for a small, but significant difference of 11-13 minutes/night in favour of auto-adjusting PAP in all(246) or only cross-over studies.(177) The authors however note that there is little evidence for the use of autoPAP in unselected patients. Additionally, only about 20% of the reviewed papers were clearly described as double blind studies.(177)

However, there is some evidence that pressure modifications might be beneficial in certain subgroup e.g. less compliant patients or those with a poor titration experience.(247-249) A recent pilot study investigated adherence in 51 individuals with poor initial experience with the CPAP machine who were then provided with either CPAP or an auto-bilevel machine. Although not significantly different, a higher percentage of individuals using the machine for more than 4hrs

per night were in the bi-level group compared with the fixed pressure group (62% vs. 54%). The authors attributed this null finding to a lack of power.(247)

1.3.3.1.2.2.2 Adverse Effects

Adverse effects from the CPAP machine may include dry mouth, blocked nose, skin irritation, mask leaks or frequent awakenings, to name a few. There is conflicting evidence on the relationship between side effects and adherence. The majority of studies have indicated no relationship, with reports of side effects also occurring in adherent patients(204). Interestingly, some studies indicate that specific or clusters of side effects might negatively impact on CPAP use,(182, 203, 204, 236, 250) others also argue that side effects might be more often reported in certain subgroups (Personality Type D).(251). These studies however often fail to control for these symptoms as they occur before treatment.(252, 253) One prospective study of 385 OSA patients treated with CPAP reported that some symptoms were present before the initiation of CPAP and actually improved with effective CPAP use [throat, mouth and nose dryness and nasal stuffiness]; rhinorrhoea was the only side effect that distinguished users from non-users (those who had abandoned CPAP) at 1 year(252)

Reductions in adherence rates, as a result of claustrophobic reactions to wearing a mask, have been reported in the literature.(181, 254) Only one study to date has identified the impact of a ‘traitlike’ claustrophobic tendency on CPAP use. Chasens and colleagues identified individuals with higher pre-treatment claustrophobia were almost twice as likely to use their machines for less than 2hrs/night than those with a lower claustrophobia score.(254) Interventions to reduce claustrophobic reactions to the mask have been successful as outlined below.

1.3.3.1.2.2.3 Mask Interface

Increased attention has been devoted to improvements in the CPAP mask interface to make this treatment more acceptable for patients. However only a few studies have investigated the effect on adherence in randomised controlled environment, which leads to uncertain conclusions as to whether less obtrusive interfaces (e.g.

nasal pillows/nasal masks) are associated with higher adherence than their more obtrusive counterparts (e.g. oral/face masks).(255) Equally, limited data is available for humidifiers. The addition of humidification may reduce the presence of nasal side effects such as rhinorrhoea(256), however there is limited evidence to suggest that this improves CPAP use, and differences are often minor.(177, 256)

1.3.3.1.2.2.4 CPAP pressure titration

Variations of the titration method are generally not associated with any differences in subsequent CPAP use. Comparisons include lab versus home(257-260), manual vs. automatic vs. algorithm based(244, 258, 261-264), attended versus unattended in-lab(265), split night vs. full night(266-269), one versus two full night titrations(270), or variations of more than two methods(270-274). What might be more predictive of adherence is the actual quality of the titration night, especially when evaluating patients' sleep quality. A qualitative study described those with a poor titration night experience were most likely to report difficulties using a machine.(275) This finding has been corroborated with recent quantitative studies. Drake and colleagues reported that in 71 OSA patients, who had improved sleep quality during the titration study compared to the diagnostic overnight stay, used the machine for approximately 2 hrs more per night during the first 1½ months. This result remained significant after controlling for OSA severity as a potential moderator.(276) A number of studies have attempted to prospectively improve the sleep quality of the titration night. Following support for the use of a sedative hypnotic in a retrospective study and its effect on titration night sleep quality and subsequent adherence,(277) Lettieri and colleagues conducted a prospective randomised control trial in 98 individuals with OSA comparing eszopiclone or placebo pill during the titration night.(278) Improvements in sleep quality and a 1 hr increase in CPAP use was reported on days used at 4-6 weeks in the active treatment arm compared to the placebo. In contrast, one study tested the effects of an auto-PAP machine "pre-treatment" for 2-months prior to the lab titration. Significantly increased sleep efficiency was found during the titration night compared to the control group (no pre-treatment prior to titration night), however

at 9- months follow-up there was no significant difference in adherence between the groups (5.1 ± 0.5 and 4.9 ± 0.4 hrs/night).(279)

1.3.3.1.2.3 Disorder-Specific

1.3.3.1.2.3.1 Disease severity (AHI, ODI) and BMI

Positive relationships have been reported between OSA severity and CPAP adherence, these relations are however weak and a large proportion of studies show no relationship. Furthermore, there is no consensus on the appropriate measure of disease severity, some studies report a relationship with the AHI(240, 280, 281), some with the ODI(241) and others with the RDI(29), which is known to underestimate severity as it represents the events per hour of recording (thus including nightly wake time). Some studies have indicated that greater BMI is related to increased CPAP use, but again these relationships are weak.

1.3.3.1.2.3.2 Symptomatic Severity (Baseline ESS)

Higher rates of daytime somnolence at baseline have been associated with adherence in some studies, yet improvements in sleepiness post treatment might be a better predictor. Symptomatic improvement (reduction in sleepiness, daytime functioning, and health related quality of life) post treatment is associated with increased CPAP use. Higher CPAP use might be a result of reductions in functional symptoms; however, the reverse might also be true: resulting from a dose-response relationship, whereby increased use leads to increased improvements in symptoms.(59) There is tentative evidence to suggest both scenarios are equally valid. In the attempt to define determinants of CPAP use in South Florida Hispanic veterans, Wallace and colleagues reported that normalisation of the Epworth Sleepiness Score at week 1 (<10) remained a significant predictor of CPAP use at 1 month, even when controlling for week 1 CPAP use.(236) Additionally, Weaver and colleagues reported only 30% and 50% achieved normal values on the multiple sleep latency test (MSLT) and Functional Outcomes of Sleep Questionnaires (FOSQ) respectively despite using the machine more than 7hr per night.(57) Antic et al. reported similar effects: one in 5 patients did not obtain normal sleepiness values

(Epworth<10) at follow-up despite adequate use,(58) thus not all of the improvement can be attributable to adherence. These two dose-response studies argue against a clear path from increased use to symptomatic improvement and together with Wallace's study (partly) support the direction that symptomatic improvement→ increased use.

Support for the alternative direction is provided by a recently published study(1), which describes how part of the does-response relationship is a result of an expectation of benefit. Improvements as a result of increased use were witnessed in the placebo arm, where no therapeutic pressure was provided, supporting the direction increased use→ increased improvement. However, these effects were also observed in the therapeutic arm (and to a larger extent), suggesting a possible interactive feedback loop, whereby increased use leads to improvements, which further motivates CPAP use.

1.3.3.1.2.3.3 Nasal Resistance

There is emerging evidence to support the association between upper airway resistance and CPAP adherence and this link is potentially two-fold.(282). A narrow upper airway may increase the perception of increased breathing effort and ultimately decrease CPAP use. Additionally, a narrow nasal passage and increased resistance might increase the frequency of mouth breathing and ultimately the possibility of mask leaks. However, the relationship might largely be related to initial acceptance rather than long-term CPAP use.(283-285) Interestingly, studies have indicated that nasal resistance/stuffiness measured subjectively does not predict CPAP adherence/acceptance(252, 253, 282, 284) even in patients where objective resistance does.(282, 284)

1.3.3.1.3 Psychological

It is clear from the above overview, that patient and treatment specific biomedical factors measured alone are relatively poor predictors of CPAP adherence. Thus, turning the attention also towards psychological and social factors might prove beneficial in our overall understanding of the factors that influence adherence.

1.3.3.1.3.1 Cognitive/Motivational

1.3.3.1.3.1.1 Knowledge

It is unclear whether increased knowledge of the disorder and its treatment, when measured as an independent variable, procures improvements in adherence. When knowledge is considered in combination with other variables of Bandura's social cognitive theory (self-efficacy, outcome expectancy, and social support) the model explains between 11-40% of the variance in CPAP use.(286, 287) A recent meta-analysis indicated that educational interventions do not generally produce improvements in CPAP use when contrasted with usual care, as outlined below.(288) The failure to measure post-intervention knowledge precludes the evaluation of the direct relationship between knowledge and CPAP use in these comparison studies.

Table 6: Variables influencing CPAP adherence

Variables	Impact on Adherence	References
Biomedical		
Demographics/Patient Specific		
Age (Ref=increased age)	Increase	Woehrle 2011(243), Fuchs 2010[+](289), Means 2010(290), Amfilochiou 2009(291), Collen 2009(277), Simon-Tuval 2009[+](292), Budhiraja 2007(199), Li 2005(282), Scharf 2004(29), Sin 2002(195), McArdle 1999(196)
	Decrease	Janson 2000[+](293)
	No Difference	Alves 2012(201), Pieh 2012(294), Wallace 2012(236), Bakker 2011(244), Gagnadoux 2011(280), Galetke 2011(295), Letteri 2011(273), Sawyer 2011,(296) Ye 2011(297), Kohler 2010§[+](241), Nguyen 2010(235), Baron 2009(298), So 2009[+](285), Sopkova 2009(299), Poulet 2009§(300), Olsen 2008(301), Wolkove 2008[+](192), Yetkin 2008[+](302), Sugiura 2007[+](283), Morris 2006[+](284), Aloia 2005(237), Chasens 2005(254), Lewis 2004(303), Lloberes 2004(281); Wild 2004(304), Holland 2003(305), Ball 2001[+](306), McFayden 2001(307), Pelletier-Fleury 2001§(240); Sage 2001(308), Noseda 2000(309), Sanders 2000(268), Pepin 1999(202) Weaver 1997(203); Edinger 1994[+](310), Kribbs 1993(181), Rauscher 1991[+](191), Waldhorn 1990[+](311), Nino-Murcia 1989[+](312)
Gender (Ref=male)	Increase	Woehrle 2011(243), Joo 2007(313), Llobreres 2004(281), Pelletier-Fleury 2001(240)
	Decrease	Amfilochiou 2009(291), Sin 2002(195)
	No Difference	Alves 2012(201), Campbell 2012(271), Stuck 2012(314) Pieh 2012(294), Wallace 2012(236), Bakker 2011(244), Gagnadoux 2011(280), Galetke 2011(295), Letteri 2011(273), Ye 2011(297), Fuchs 2010[+](289), Kohler 2010[+](241), Platt 2009(315), Poulet 2009§(300), So 2009[+](285), Sopkova 2009(299), Ye 2009(316), Olsen 2008(301), Wolkove 2008[+](192), Budhiraja 2007(199), Morris 2006[+](284), Aloia 2005(237), Chasens 2005(254), Li 2005(282), Lewis 2004§(303), Scharf 2004(29), Ball 2001[+](306), Sage 2001(308), Janson 2000[+](293), Weaver 1997(203), Meurice 1994(193), Rauscher 1991[+](191), Waldhorn 1990[+](311), Nino-Murcia 1989[+](312)
Race/Ethnicity (ref= minority)	Decrease	Campbell 2012(271), Pamidi 2012(317), Sawyer 2011(296), Ye 2011(297), Means 2010(290), Budhajara 2007(199), Joo 2007(313)
	No Difference	Bakker 2011(244), Platt 2009(315), Chasens 2005(254), Scharf 2004[+](29)
Treatment Specific		
Pressure Modifications (ref= Bi-level, C-flex, APAP)	Increase	Ip 2012(246), Gentina 2011(248), Smith 2009 [auto-CPAP in cross-over studies](177)
	No Difference	Powell 2012(247), Blau 2011(318), Kushida 2011(319), Bakker 2011(245), Khayat 2008(320)
Adverse Effects (ref=presence of side	Increase	Brander 1999(253), Weaver 1997(203)

effects)	Decrease	Wallace 2012(236), Ye 2011(297), Brostroem 2010(321), Kreivi 2010+(252), Baltzan 2009(250), Aloia 2007(204), Broström 2007(251), Chasens 2005(254), Lewis 2004(303), Kalan 1999+(322), Engleman 1996+(182), Engleman 1994(187), Kribbs 1993(181), Hoffstein 1992+(179)
	No Difference	Stepnowsky 2006§(287), Drake 2003(276), Russo-Mango 2001(194), Pepin 1999(202), Pepin 1995(323), Meurice 1994(193), Rauscher 1993(183), Waldhorn 1990+(311)
CPAP delivery interface (ref=less obtrusive)	No Difference	Ryan 2011(324), Nguyen 2010(235), Li 2005(282)
	Unclear (meta-analysis)	Chai 2006(255),
Humidifiers	No Difference	Ryan 2009(256)
	Unclear (meta-analysis)	Smith 2009(177)
Pressure (ref=higher pressure)	Increase	Janson 2000+(293), Nino-Murcia 1989+(312), Kohler 2010+(241)
	Decrease	Wild 2004(304), Pelletier-Fleury 2001(240)
	No Difference	Pieh 2012(294), Wallace 2012(236), Bakker 2011(244), Kreivi 2010+(252), Amfilochiou 2009(291), So 2009+(285), Sopkova 2009(299), Berry 2008(262), Olsen 2008(301), Wolkove 2008+(192), Wells 2007(325), Morris 2006+(284), Stepnowsky 2006(287), Aloia 2005(237), Lewis 2004(303), Holland 2003(305), Stepnowsky 2002§(286), Ball 2001+(306), Russo-Mango 2001(194), Sage 2001(308), Pepin 1999(202), Meslier 1998(200), Weaver 1997(203) Kribbs 1993(181), Rauscher 1993(183), Waldhorn 1990+(311),
(Sleep) quality of titration night (ref=better quality)	Increase	Collen 2009(277), Lettieri 2009(278), Drake 2003(276), Lewis 2004(303)
	No Difference	Suzuki 2007(279)
Titration method (ref=traditional in-lab full night titration)	Increase	Means 2004(265),
	Decrease	Rosen 2012(258), Mulgrew 2007(260)
	No Difference	Campbell 2012(271), Pamidi 2012(317), Wallace 2012(236), Kushida 2011(319), Bakker 2011(244), Skomro 2010(259), Letteri 2011(273), Collen 2010(266), McArdle 2010(272), Platt 2009(315), Berry 2008(262), Kaplan 2007+(270), Cross 2006(257), West 2006(264), Hukins 2005(263), Lloberes 2004(274), Masa 2004(261), McArdle 2000(269), Sanders 2000(268), Strollo 1996(267)
Disorder Specific		
Disease severity (AHI, RDI ODI, ref=increased severity)	Increase	Tarasiuk 2012(326), Gagnadoux 2011(280), Galetke 2011(295), Kohler 2010+(241), Kreivi 2010+(252), Means 2010(290), Simon-Tuval 2009+(292), So 2009+(285), Yetkin 2008+(302), Sugiura 2007+(283), Bizieux-Thamin 2005(327), Lloberes 2004(281), Scharf 2004(29), Wild 2004(304), Drake 2003(276), McFayden 2001(307), Pelletier-Fleury 2001(240), Janson 2000+(293), McArdle 1999(196), Meurice 1994(193), Reeves-Hoche 1994(184), Rauscher 1991(191), Nino-Murcia 1989+(312)

		No Difference	Alves 2012(201), Campbell 2012(271), Pieh 2012(294), Wallace 2012(236), Bakker 2011(244), Letteri 2011(273), Sawyer 2011(296), Ye 2011(297), Nguyen 2010(235), Amfilouchi 2009(291), Baron 2009(298), Platt 2009(315), Poulet 2009(300), Sopkova 2009(299), Berry 2008(262), Olsen 2008(301), Wolkove 2008[+](192), Budhiraja 2007(199), Kaplan 2007[+](270), Morris 2006[+](284), Aloia 2005(237), Chasens 2005(254), Li 2005(282), Lewis 2004(303), Holland 2003(305), Sin 2002(195), Ball 2001(306), Russo-Mango 2001(194), Sage 2001(308), Noseda 2000(309), Sanders 2000(268), Pepin 1999(202), Weaver 1997(203), Engleman 1996[+](182), Edinger 1994[+](310), Engleman 1994(187), Kribbs 1993(181), Rauscher 1993(183), Hoffstein 1992[+](179), Waldhorn 1990[+](311)
	Body Mass Index (ref= higher BMI)	Increase	Gagnadoux 2011(280), Kreivi 2010[+](252), Baron 2009(298), Wild 2004(304), Pelletier-Fleury 2001(240), McArdle 1999(196), Nino-Murcia 1989[+](312)
		No Difference	Alves 2012(201), Pieh 2012(294), Wallace 2012(236), Galetke 2011(295), Letteri 2011(273), Ye 2011(297), Nguyen 2010(235), Amfilochiou 2009(291), Platt 2009(315), Poulet 2009(300), Simon-Tuval 2009[+](292), So 2009[+](285), Sopkova 2009(299), Yetkin 2008[+](302), Berry 2008(262), Olsen 2008(301), Sugiura 2007§[+](283), Morris 2006[+](284), Aloia 2005(237), Chasens 2005(254), Li 2005(282), Lewis 2004(303), Scharf 2004(29), Holland 2003(305), Sin 2002(195), Russo-Mango 2001(194), Janson 2000§[+](293), Noseda 2000(309), Pepin 1999(202), Weaver 1997(203), Edinger 1994[+](310), Engleman 1994(187), Reeves-Hoche 1994(184), Kribbs 1993(181), Rauscher 1991[+](191)
	Symptomatic Severity (ref=increased e.g. daytime sleepiness or functioning)	Increase	Wolkove 2008(192), Yetkin 2008[+](302), Lloberes 2004(281), , Scharf 2004(29), Wild 2004(304) Holland 2003(305), Pelletier-Fleury 2001(240), Janson 2000[+](293), McArdle 1999(196), Engleman 1996[+](182), Reeves-Hoche 1994(184), W Rauscher 1991[+](191) Waldhorn 1990[+](311)
		Decrease	Edinger 1994[+](310)
		No Difference	Pieh 2012(294), Wallace 2012(236), Bakker 2011(244), Gagnadoux 2011(280) Lettieri 2011(273), Platt 2011(315), Sawyer 2011(296), Galetke 2011(295), Ye 2011§(297), Kohler 2010[+](241), Kreivi 2010[+](252), Nguyen 2010(235), Amfilouchil 2009(291), Baron 2009(298), Poulet 2009(300), Simon-Tuval 2009[+](292), So 2009[+](285), Sopkova 2009(299), Berry 2008(262), Olsen 2008§(301), Budhiraja 2007(199) Kaplan 2007[+](270), Sugiura 2007[+](283), Morris 2006[+](284), Aloia 2005(237), Lewis 2004(303), Stepnowsky 2002§(286), McFayden 2001(307), Sage 2001(308), Sanders 2000(268) Weaver 1997(203), Engleman 1994(187), Kribbs 1993(181), Rauscher 1993(183),
	Nasal Resistance/Upper Airway Obstruction (ref=increased resistance)	Decrease	So 2009[+](285), Amfilochiou 2009(291), Sugiura 2007[+](283), Morris 2006[+](284), Li 2005(282), Nakata 2005(328)
		No Difference	Kreivi 2010[+](252), Chasens 2005(254), Holland 2003(305), Brander 1999(253),
	Symptomatic Improvement (ref=increased improvement)	Increase	Crawford 2012(1), Weaver 2007(57), Wallace 2012(236), Antic 2011(58), Kreivi 2010[+](252), Simon-Tuval 2009[+](292), Wells 2007(325), Lewis 2004(303), Holland 2003(305), Sin 2002(195), Ball 2001[+](306) McFayden 2001(307), Kingshott 2000(59), Rosenthal 2000(238), Pepin 1999(202), Meslier 1998(200), Engleman 1996[+](182), Meurice 1994(193), Rauscher 1993(183), Kribbs 1993(181), Hoffstein 1992[+](179)
		No Difference	Amfilochiou 2009(291), Kaplan 2007[+](270), Lloberes 2004(281), Bakker(244), Engleman 1994(187),
Psychological			
	Cognitive/Motivational		

Knowledge (ref=increased knowledge)	Increase	Trupp 2011 [negatively framed information](239), Fuchs 2010+(289), Ball 2001+(306), Likar 1997(329)
	No Difference	Smith 2009 [brief education session](288), Golay 2006(330), Stepnowsky 2006(287), Stepnowsky 2002(286),
Beliefs about OSA and CPAP (ref=maladaptive beliefs)	Decrease	Poulet 2009[total score from belief scale(331)](300)
Health Value (ref=higher health value)	Increase	Wild 2004(304)
Locus of Control	No Difference	Wild 2004(304)
Perceived risk of OSA (ref=higher perceived risk)	Increase Decrease No Difference	Baron 2009(298), Olsen 2008(301), Wallace 2012(236), Simon-Tuval 2009+(292), Sage 2001(308)
Outcome Expectancies (ref=expecting increased improvements)	Increase	Olsen 2008(301), Sage 2001(308)
	No Difference	Wallace 2012§(236), Baron 2009(298), Stepnowsky 2006(287), Stepnowsky 2002(286),
Perceived barriers (ref=increased barriers)	Decrease	Sage 2001(308)
Cues to action	No Difference	Sage 2001(308)
Self-efficacy (ref=increase)	Increase	Trupp 2011(239), Sawyer 2011(296), Stepnowsky 2006(287), Aloia 2005(237),
	No Difference	Wallace 2012§(236), Bakker 2011(244), Ye 2011§(297), Baron 2009(298), Olsen 2008(301), Wild 2004(304), Stepnowsky 2002(286) Sage 2001§(308)
Readiness to change (ref=increased)	No Difference	Aloia 2005§(237)
Decisional Balance (ref=pros outweigh cons)	Increase	Stepnowsky 2006(287), Stepnowsky 2002(286)
	No Difference	Aloia 2005§(237)
Personality (=negative trait)	Decrease	Moran 2010 [behavioural inhibition](332), Broström 2007[personality type d](251), Edinger 1994+[MMPI depression](310)
	No Difference	Wallace 2012(236), Trupp 2011(239), Drake 2003(276)
Coping Style (=active coping style)	Increase	Stepnowsky 2002,(333)
	No Difference	Moran 2010(332),

	Psychological Co-morbidities		
	Insomnia (=ref increased insomnia severity)	Decrease	Pieh 2012(294), Wallace 2012(236), Wickwire 2010 [sleep maintenance problems only](334)
		No Difference	Nguyen 2010(235),
	Claustrophobia (ref=presense of claustrophobic tendency)	Decrease	Chasens 2005(254)
	Depression/Anxiety (symptoms or disorder) or other mental disorder (ref=presense of symptoms/disorder)	Decrease	Means 2010(290), Kjelsberg 2005+(335),
		No Difference	Pieh 2012(294), Wallace 2012(236), Gagnadoux 2011(280), Poulet 2009(300), Olsen 2008(301), Wells 2007(325), Lewis 2004(303), Stepnowsky 2002(333), Russo-Mango 2001(194),
Social			
	Socio-economic status (ref=higher SES)	Increase	Alves 2012(201), Tarasiuk 2012(326), Bakker 2011(244), Simon-Tuval 2009+(292), Platt 2009(315), Kribbs 1993(181), Nino-Muricia1989(312),
		No Difference	Wallace 2012(236), Gagnadoux 2011(280), Galetke 2011(295), Ye 2011(297), Campbell 2010§(271), Amfilouchi 2009(291), Poulet 2009(300), Scharf 2004(29), Weaver 1997(203),
	Marital Status (ref=living alone)	Decrease	Gagnadoux 2011(280),
		No Difference	Wallace 2012(236), Ye 2011§(297), Platt 2009§(315), Poulet 2009(300), Olsen 2008(301), Kaplan 2007+(270), Lewis 2004§(303), Wild 2004(304) Russo-Mango 2001(194),Weaver 1997(203), Kribbs 1993(181),
	Bed-sharing (ref=sleeping alone)	Increase	Simon-Tuval 2009+(292),
		Decrease	Cartwright 2008(336)
	Partner's sleep quality (ref=improvement)	Increase	McArdle 2001(337)
	Relationship Quality (ref=greater quality)	Increase	Baron 2009[conflict but not support subscale related to adherence](298),
		No difference	McFayden 2001(307),
	Pressure from Spouse (ref=increased pressure	Decrease	Baron 2011(338),
	Support (ref=increased support)		
	General	No Difference	Stepnowsky 2006(287), Stepnowsky 2002(286)
	Spouse/Close social relationships	Increase	Baron 2011+(338), Simon-Tuval 2009+(292),
	Therapist	Increase	Russo-Mango 2001(194), Smith 2009(288),

§ becomes non-significant when controlling for other independent variables. + compared to subjective adherence only, or attendance rates/acceptance/termination of CPAP as a primary outcome, AHI=Apnea Hypopnea Index, BMI=Body Mass Index, CPAP=Continuous Positive Airway Pressure, MMPI= Minnesota Multiphasic Personality Inventory, ODI= Oxygen Desaturation Index, RDI= Respiratory Disturbance Index, SES=Socio-economic status

1.3.3.1.3.1.2 Beliefs about the disorder and treatment

A number of beliefs about the sleep disorder and the treatment have been established as predictors of CPAP use. An exciting trend in the literature is the adoption of social cognitive and stage models to explain CPAP adherence, ranging from the health belief model, social cognitive theory, the transtheoretical model and Wallston's health locus of control and explaining about 11-58% of the variance in CPAP use.

Wild and colleagues found that components of the health locus of control model along with AHI, CPAP pressure BMI and daytime sleepiness, could explain 24 % of the variance in CPAP use.(304) Attribution of control to oneself, less belief in control of others and greater value of health were included in the final model, however only the latter variable was a significant independent predictor of CPAP use. Self-efficacy and belief that the condition is influenced only by chance had no significant impact on adherence in the 119 OSA patients. This null finding might be explained by the measurement of self-efficacy prior to initiation of treatment, as reported in other studies(237, 244, 286, 296, 298, 300, 304). When measured post CPAP experience this variable becomes a significant predictor of CPAP use,(236, 237, 287, 296, 333) however not after controlling for prior objective CPAP use.(236, 237) Self-efficacy features in a number of social cognitive and stage models, most notably Bandura's social cognitive theory (SCT)(339, 340), which has been rigorously tested by one particular working group.(237, 286, 287) Stepnowsky and colleagues(286) found that outcome expectancy, self-efficacy, social support and knowledge measured at 1 week and 1 month accounted for 26% and 40% of the variance in CPAP use at 1 month respectively. In similar studies they replicated these findings in 98 OSA individual's long term use of 6-months(237) as well as 58 experienced CPAP users.(287)

Apart from the SCT, these studies also investigated the predictive validity of the transtheoretical model (TTM). This dynamic stage-based model is further outlined below, but briefly it encompasses four components; the stage of change, processes of change, the decisional balance (pros vs. cons) and self-efficacy/temptation. In the short-term follow-up study, TTM variables decisional balance, processes of change and stage of change were measured and explained 17% and 33% when measured at week 1 and 1 month, yet baseline values were

not predictive of CPAP use.(286) The only component in the cross-sectional study was the decisional balance and together with side effects and CPAP pressure, it explained 47% of objective CPAP use.(287) The long-term follow-up study measured readiness to change and the decisional balance and together with self-efficacy explained between 23-58% of the variance at week 1 and months 3 and 6. However, this model alone did not significantly succeed beyond the predictability of prior objective CPAP use.(237) Unfortunately, these studies failed to include all four components of the TTM within the same model and thus it is difficult to evaluate the TTM **model** from these studies.

Another social cognitive model that has been investigated is the health belief model (HBM). Sage et al. 2001(308) and Olsen et al. 2008(301) examined HBM variables adherences post and pre CPAP initiation respectively. In a sample of 40 naïve CPAP users, Sage and colleagues found a univariate association between use and self-efficacy and only barriers to and benefits of adherence were independent predictors of CPAP use after one month. In this sample, HBM variables were assessed with a thematically organised scale against the components of the model. The aim of Olsen's study was to replicate these findings when predictor variables were assessed prior to any experience of CPAP use. In their sample of 77 newly diagnosed OSA patients, they found benefits; severity and susceptibility and self-efficacy explained 21.8% of the variance in CPAP use at 4 months. Interestingly, this study made use of already validated sleep questionnaires, such as the Functional Outcomes of Sleep Questionnaire (FOSQ, 341) and the Self-Efficacy Measure for Sleep Apnea (342) as equivalents to the constructs of the HBM. No validated sleep apnoea specific questionnaires for the constructs 'barriers' 'cues to action' 'health motivation' or 'barriers' existed at the time and therefore were not assessed. An additional limitation of this study was the use of the FOSQ. This scale measures severity of sleepiness-induced impairment in daytime functioning; however this does not equate to a complete theoretical overlap with the severity construct as outlined in the HBM. Although sleepiness is a motive for treatment seeking behaviour in the majority of individuals diagnosed with OSA, (341) not all present with excessive daytime sleepiness.(10, 54) Severity of all symptoms, not just sleepiness needs to be assessed.

There are a number of studies that have assessed more trait-like variables such as personality and coping style. Limited information is available regarding personality measures; the inability to avoid unwanted behaviours (behavioural

inhibition system model of personality)(332) and personality type D have been associated with poorer CPAP adherence.(251). A study, which to date has predicted the largest amount of variance in CPAP adherence, used the Minnesota Multiphasic Personality Index.(310)The final logistic regression model included Body Mass Index, daytime sleepiness, nocturnal sleep quality, and two of the MMPI subscales: depression and hypochondriasis, explaining 63% of the variance, however only depression and less pre-treatment sleepiness were univariate predictors of better subjective CPAP use. The final model could correctly identify 75% of 8 non-compliers and 90% of the 20 compliers after 6 months.

In summary, these cognitive/motivational variables when measured after some exposure, even merely a brief education session(296) are predictive of adherence, yet rarely when measured at baseline, which suggests that at least one form of exposure to CPAP is needed for the patient to formalise these beliefs. Studies that are based on social cognitive frameworks are usually superior in predicting CPAP use, and provide a holistic framework for interventions

1.3.3.1.3.2 Psychological Co-morbidities

The co-morbidity between insomnia and OSA is high, with prevalence of insomnia in those who present to a sleep clinic for assessment of possible sleep related breathing disorders ranging between approx. 40-50%.(343, 344) However the possible impact of insomnia on CPAP adherence is relatively under-researched. One of the first studies to examine the impact of insomnia on CPAP adherence found Insomnia Severity (ISI) total score and insomnia symptoms (first three items of ISI), were unrelated to attrition rates or CPAP adherence at 6-months, using data mining techniques in 148 CPAP users.(235)

In contrast, a recent study in Hispanic veterans found that in the 65 completed 1-week follow-up and 59 who attended also the 1-month follow-up, lower ISI score was the only independent predictor of increased CPAP use at 1 week (65 CPAP users), but not at one month (59 CPAP users), where only increased 7-day adherence and daytime sleepiness normalization (ESS<10) were independent predictors.(236) In addition, one chart review of 232 OSA patients, who had been prescribed CPAP, and had provided information on possible insomnia symptoms, revealed that sleep maintenance symptoms were predictive

of average CPAP use and adherent status (≥4hrs use on 70% of nights) on average 4 ½ months after titration, even after controlling for age and gender.(334) Unfortunately this study only assessed insomnia symptoms to classify individuals into one of three groups: 'sleep onset insomnia', 'sleep maintenance insomnia' and 'early morning awakenings', and thus not considering important symptoms necessary for an insomnia diagnosis: 1) dissatisfaction with amount of sleep 2) concomitant daytime dysfunction. The relationship with maintenance problems only is potentially explained by the timings of these awakenings coinciding with time of impaired rational decision making (i.e., when woken from sleep).

Other mental health symptoms or disorders such as anxiety and depression may impact on adherence to CPAP, however there are conflicting results as can be seen in Table 6. The difficulty with these studies is that higher rates on these scales might be a result of non-adherence per se, in addition to, or rather than, a predictor of poorer adherence. Further prospective studies are needed to investigate this further.

1.3.3.1.4 Social

Obstructive sleep apnoea and the treatment of choice affect the patient's social surroundings on a variety of levels. Not only do the nocturnal symptoms affect the partner's sleep, but also sleepiness and irritability can put a strain on family life. The eventual presentation of CPAP to family life may be associated with embarrassment as a result of taking on a sick role and changes to the nightly routine, which may also include the re-introduction of bed sharing. Thus inevitably, the condition and its treatment affect not only the patient, but also the bed partner and family. Social influences on CPAP adherence have only recently been focus of increased attention.

1.3.3.1.4.1 Socio-demographic factors

There is uncertainty about the effect of socio-economic status (SES) on CPAP adherence. The majority of studies outlined in Table 6 differ in the definition of SES, which may include all or only one of these three variables: income, education and occupation. Some studies have indicated that lower socio-economic status is associated with a decrease in CPAP acceptance and use, even in multivariate analysis controlling for other variables.(244, 292) Another socio-demographic factor, marital status (living alone versus living with a partner/spouse) does generally not relate to adherence, see Table 6, which is

perhaps surprising, considering a live-in partner might provided augmented support. On the contrary, spousal pressure to adhere might effectively reduce adherence rates; certainly, when self-referred adherence tends to be increased compared to partner-referred.(345)

1.3.3.1.4.2 Social involvement and relationship quality

Baron and colleagues have investigated exactly this, by measuring three different types of spousal involvement: pressure, support and collaboration.(298) In 31 couples the authors determined the intricate, directional relationship between support and CPAP adherence using a day-to-day study design. Whilst controlling for previous day adherence and wife involvement, nightly CPAP use was predicted by previous day wife support, however only in high disease severity patients. Interestingly, spousal collaboration was increased after nights with poor adherence, and the authors relate this to possible sleep disruption caused by the re-emergence of nocturnal symptoms (e.g. snoring) with non-CPAP use. Spousal pressure was not related to daily measures, however it did predict poorer objective adherence at 3 months. The caveat of this study was that daily adherence was measured subjectively.

Apart from support, other variables that have been investigated are relationship quality; the partner's sleep quality and whether or not the patient is sharing a bed with the partner, see Table 6. Cartwright investigated the co-sleeping habits in 10 male OSA patients during a 2-week period post CPAP initiation.(336) A strong correlation of r=.74 was reported between number of co-sleeps and adherence rates. Co-sleeping was also related to the wife's arousal index on both the pre- and post-treatment polysomnography, which might indicate that partners who are not co-sleeping are in fear of being woken by the CPAP machine. Whether these partners are less accepting of the CPAP machine remains to be established. The 2-week period was not sufficient to determine whether changes to sleeping habits long-term predict CPAP use.

In addition to partner/close relationship support, therapist support may play a vital role in CPAP adherence. Certainly qualitative studies have revealed that patients' behaviour is encouraged by the supportive environment at the sleep centre and/or CPAP distributor, see chapter 3. There are also a number of

intervention studies comparing intensive therapist and/or nurse support to treatment as usual care, see below.

In summary, it is evident that the social aspect of CPAP adherence has been largely neglected by the literature. However, the studies that have focused on this are revealing interesting angles of the multifaceted relationship between CPAP adherence and social interactions.

1.3.3.2 Predictors of Adherence to CBT-I

As evident from Table 5 only eight studies have examined potential predictors of behavioural and cognitive components of CBT-I. Of all predictor variables presented in this table, four common variables seem to emerge in a number of studies: depressive symptoms,(214, 215, 221) pre-treatment symptom severity(208, 232), readiness/motivation for change(217, 223) and belief in the ability to adhere to the treatment programme.(208, 223, 231)

Although two studies indicated no relationship with depressive symptoms,(217, 232) this symptom has been linked to poorer adherence in others. In the largest study of 301 clinical outpatients undergoing CBT-I and who had completed the Beck Depression Inventory (BDI) prior to treatment, Manber and colleagues reported that those with increased depressive symptoms (BDI score≥ 14) had increased difficulty adhering to the behavioural components, than those with lower BDI scores (<14)(215). Significant differences were found for rising time and restricting time in bed, possibly indicating that those with increased levels of depression find it more difficult to get out of bed to start the day. Unfortunately employment status was not measured in this sample, a factor that might have been a moderator (or even mediator) of this relationship. There was a trend for a significant difference between the two groups and adherence to cognitive components. Similar relationships have been reported in other studies. For example, Vincent and colleagues in 2003(214) reported that therapist reported patient adherence was reduced when dysthymia was present. A crucial limitation of this study was the assessment of a co-morbid dysthymia (using the Structured Clinical Interview for DSM-III-R anxiety and mood disorders (SCID)), which was carried out by the same psychologist who ran the groups and provided therapist rated patient adherence. Using structural equation modelling,(221) McChargue and colleagues identified that a increase in depressive symptoms, as assessed with the Hospital Anxiety and Depression Scale (HADS), was predictive of a decrease in adherence to a modified sleep restriction component in cancer patients. Additionally, these authors found a decrease in sleep disturbance was associated with a reduction in adherence to relaxation strategies, potentially as a result of the implementation becoming redundant with perceived improvements. The authors however do not specify whether baseline symptom severity was controlled for: potentially those with higher baseline sleep disturbance have more room for improvement and are as a

result more adherent initially, becoming less adherent with witnessed improvements.

The relationship between adherence and pre-treatment sleep quality and sleepiness severity has been reported in other studies with differential effects. Morgan et al. reported those with poorer sleep quality at baseline (measured with the PSQI) were more likely to attend all CBT-I sessions.(232) A workgroup from Canada on the other hand, described increased sleepiness at baseline (measured with the Epworth Sleepiness Scale, ESS) was associated with *poorer* adherence (less consistency in wake-up time).(208) Of note, Vincent et al's measure of **wake-up** time should not truly be considered as a measure of adherence, as this variable is out with the individual's control. Apart from differences in study design, treatment components and adherence measures, these differential effects might be explained by the finding that individuals with insomnia rarely complain of sleepiness as a marker of increased hyperarousal.(346) Pretreatment sleepiness as defined in Vincent et al.'s study should thus not be considered a symptom of insomnia that improves throughout the course of treatment, but a symptom that potentially increases as a side effect of treatment implementation.(see 139) Alternatively, one could make the tentative suggestion that increased pre-treatment sleepiness is a marker of less-severe hyperarousal. In contrast higher levels of fatigue at baseline as a symptom of insomnia might be predictive of insomnia, as has been indicated in a recent study in breast cancer patients with co-morbid insomnia.(217)

Readiness/motivation to change has been a large predictor of various health behaviours such as smoking, substance misuse and exercise. Two recent studies have examined this variable in relation to CBT-I adherence.(217, 223) One study measured readiness to engage in an online cognitive behavioural programme and stated that individuals who were more contemplative (thinking about change) were more likely to engage in sleep hygiene instructions.(223) Corroborating the importance of motivation, a study in breast cancer patients with co-morbid insomnia and higher baseline motivation scores were more likely to adhere to their prescribed rising time and time in bed.(217) A detailed description of these two studies is presented in chapter 6, which examines the use of the transtheoretical model for predicting adherence.

The last variable seemingly emerging as a common denominator in various studies, is the belief in the ability to engage in treatment components, often

termed self-efficacy or perceived behavioural control. Although these constructs are often considered separate entities, they both describe the belief in the capabilities of carrying out the behaviour of interest.(347) In 2003 as part of a larger trial, Bouchard and colleagues examined self-efficacy at a general, task specific and self-regulatory (when obstacles arise) level and reported no (.17) to strong (.67) correlations between higher levels of self-efficacy (at all levels) across treatment. There was a U-shaped curve with the strongest correlations reported halfway through the 8-week treatment (weeks 3-5), this decrease in correlations after week 5 authors attributed to increases in adherence and the inability of the conservative adherence measure to distinguish between good and excellent adherers. Generally though, this study provided preliminary evidence that individuals, who believe in their abilities to implement the behaviour are more likely to do so. One working group in two separate studies has reported a similar finding.(208, 223) In 2010, Hebert et al. explored the use of components from the Transtheoretical Model (TTM, see description below) and the Theory of Planned Behaviour in explaining adherence and attrition rates to an online CBT-I program.(223). When measured prior to any exposure to treatment, perceived behavioural control was strongly correlated (r=.5) with adherence (defined as the percentage of participants using homework for at least 4 nights of the week). Unfortunately, the scale assessed the perception of participating in the online program, rather than implementation of the components themselves e.g. “Do you think it would be easy or difficult to complete the online treatment program in the next 5 weeks?” These findings are partly supported by another study from this working group,(208) who reported perceived certainty in adhering to stimulus control and sleep restriction procedures (an item of their perceived behavioural control scale) was moderately correlated (r=.3) with consistency in wake-up time. However in the overall regression model, perceived behavioural control did not emerge as a significant predictor of adherence.

Other variables that have been associated with poorer adherence are the use of medication and perceiving fewer barriers with treatment,(208) and factors related to increased adherence are social support, intention to engage in behaviour change(223) and pre-treatment perception of CBT-I(230) Younger adults were found to be more adherent in one study,(208) however age was not a significant predictor in Morgan et al.’s study.(232)

In summary, three factors seem to emerge from the existing literature as possible strong predictors of adherence to CBT-I. These are depressive symptoms, pre-treatment symptom severity, readiness/motivation to change and self-efficacy/perceived behavioural control. Undoubtedly, though there is considerable scope to investigate further predictors of adherence, and research clearly lags behind that on CPAP adherence literature.

1.3.4 Interventions to Improve Adherence

Establishing predictors of adherence can benefit the development of interventions, which in turn may improve outcome and subsequently increase the effectiveness of a treatment (over and above treatment efficacy).(226) A recent Cochrane review of interventions for medication adherence concluded that there was some evidence to suggest simple interventions produced improvements short-term, however for long-term care (e.g. diabetes, hypertension) most improvements were reported in studies that included complex interventions with combinations of educational, psychological and/or behavioural components. Surprisingly, even the most effective interventions did not lead to a meaningful improvement in both adherence and outcome. However, the authors note that these negative conclusions might be partly explained by poorly defined adherence measures, some poorly designed interventions, underpowered studies and stringent inclusion criteria for studies reporting *both* adherence and outcome.(348)

1.3.4.1 Interventions to improve CPAP adherence

1.3.4.1.1 Technological

Recent advancements in technology have generated several improvements to CPAP treatment from the original presentation in 1981. These include changes to the mask interface, the addition of humidifiers and pressure modifications (Auto, Bi-Level, C-Flex and various combinations of these three modalities). As outlined in Table 6 these adaptations generally do not procure improvements in CPAP adherence, and this is perhaps unsurprising, because they are mainly designed to reduce side effects, which again are not clearly linked to changes in CPAP use. As described above, three recent meta-analyses(177, 245, 246) of technological interventions revealed no evidence for increased adherence in any

modality except for a small, but significant difference of 11-13 minutes in favour of auto-adjusting PAP in all(246) or only cross-over studies.(177) There is however some evidence for these pressure modifications in unselected patients, e.g., those who require higher CPAP pressure, are non-adherent or had poor first experience with CPAP might benefit from these modes as described above.(177, 247, 248)

One side effect that has consistently been linked to poorer adherence is claustrophobic reactions to the mask(181, 254) and interventions to reduce these have been successful. Following a case report(349), Edinger's group retrospectively examined the effects of gradual desensitisation therapy in 13 patients who had reported claustrophobia. Post-intervention, in which patients were gradually introduced to the machine based on individualised hierarchy of steps to acclimatise to the devise, CPAP was used on average 4.4hrs (SD=0.6) on only the days implemented, which was significantly more compared to pre-intervention rates (mean 1.8 hrs, SD=0.6), indicating a large effect size of Cohen's d=1.2. Similarly large effects were reported for percentage of nights CPAP was used and mean hours of CPAP use (all nights). However only half of the patients met criteria for adequate adherence (≥4hs on 70% of nights), and these low adherence rates might relate to this being a clinical compared with a research sample, and as a result there was an of extremely large variation in time between titration and desensitisation therapy (2.6 weeks–6.2 years). The effects of gradual exposure therapy in a prospective randomised control trial for these patients, or administered preventatively for individuals with claustrophobic tendencies(254) is warranted.

1.3.4.1.2 Educational interventions

In 2009 Smith and colleagues published an updated Cochrane review examining the effect of brief one-off education sessions on subsequent adherence.(288) Three studies were selected that had machine usage as the main outcome, however the pooled difference of 0.11 hrs/night in favour of the intervention was not significant with 95% CI between -0.7 and 0.93. A similar result was reported for the four studies measuring withdrawal rates. In addition to these randomised control trials, case studies that have provided a one-off education and comparing pre to post adherence rates have reported no significant difference after an extensive education session for patient and spouse in one

study,(330) and only a small difference after group education sessions of d=0.3.(329)

A recent study examined the impact of the actual nature of the educational message. Trupp and colleagues found that adherence was significantly higher in OSA patients who received negatively framed educational messages [focused on the consequences of not using CPAP] as opposed to positively framed [focus on the benefits of using CPAP].(239) More than 4 hrs per night on at least 70% of nights was obtained by 55% of those receiving the negative message, compared to only 23% in the positively framed group.

It is important to note, however that education is not classically considered an independent predictor of behavioural adherence, but rather forms adequate circumstances under which behaviour change can occur contingent on other beliefs and attitudes (e.g. self-efficacy).(350) Thus, education is often considered integral to optimal patient care in OSA patients.(41)

1.3.4.1.3 Complex Interventions

Complex interventions such augmented support, cognitive behavioural therapy or motivational interviewing have been examined as potential interventions to improve adherence. The Cochrane review described above analysed results from ten studies and reported a significant difference of .59 hours in use in favour of augmented educational, psychological or practical support in form of follow-up home visits or telephone/internet support from either a nurse or physician.(288) With large statistical variation in these studies and despite sensitivity analyses, the authors were unable to determine whether this was a consistent effect.

Perhaps the greatest effect of any interventions tested in a randomised trial thus far have been for psychological interventions such as cognitive behavioural(351, 352) or motivational enhancement(353-355) therapy, with large effect sizes at 1 month (where reported) of d=0.5-1.1.

Richards and colleagues randomised 100 OSA patients to receive either cognitive behavioural therapy (CBT) or treatment as usual. CBT was based on Bandura's social cognitive theory and aimed to target self-efficacy, outcome expectations, social support and knowledge with two 1hr education sessions separated by 1 week. Sessions were aimed to correct distorted beliefs and encourage a positive outlook for both patients and their partner. Individuals

were also aided in the initial handling of the mask, provided relaxation techniques to deal with any anxious reactions and were shown a 15-minute video of role models. Lastly, patients were sent home with a booklet that outlined strategies for dealing with maladaptive beliefs about the CPAP machine. CPAP use and percentage of patients with more than nightly 4hr use was significantly improved in the CBT group at both 7 days and 1 month, with large effect sizes of around 1.1. Self-efficacy, social support, but not outcome expectations were significantly increased in the intervention group.(351)

Following successful support interventions via the tele-health route in significantly improving CPAP use(356, 357) Sparrow and colleagues tested the effect of motivational enhancement therapy for CPAP(358) in the context of a telephone linked communication system.(355) A total of 250 CPAP prescribed OSA patients were randomised to either an intervention group or tele-health placebo control, which providing information about other health aspects. The intervention was designed to assess patient's motivation, goals, barriers to use CPAP and self-efficacy and aimed to increase each of these aspects by using three non-judgmental and empathic strategies consistent with motivational interviewing: 1) decisional balance (although this is not strictly a MI technique, but has often been misrepresented as such(359)), e.g. "On the one hand you noticed discomfort as a problem with CPAP, but on the other hand you see the benefits to your health. Please think more about that." [online supplemental material], and 2) self-efficacy and 3) motivation rulers, with which patients rate their confidence and motivation to continue using CPAP on a scale of 1 to 10. The intervention arm used the CPAP machine 1hr more and even 2hrs more per night at 6 and 12 months respectively. Unfortunately the adherence rates were low in both arms (less than 3hrs/night on average), which might be attributed to the late introduction of the intervention, i.e. after initiation of CPAP. Interestingly, the intervention produced significant effects for self-efficacy and the decisional balance and mediation analysis revealed that 60% of the variance explained by the treatment effects was actually attributable to these psychological variables.

In summary these cognitive behavioural and motivational enhancement type interventions seem to be effective for improving CPAP use, however the literature could benefit from further and larger randomised control trials

introducing these interventions prior to CPAP titration and potentially even targeting those struggling or have low self-efficiency and motivation to change.

1.3.4.1.4 Treating co-morbidities

As outlined above and in Table 6, co-morbidities are common in this population, especially insomnia. To date no study has directly investigated the effect of treating co-morbid insomnia and how this impacts on CPAP adherence. However a number of studies provide preliminary support for this approach, by targeting aspects that may be particularly problematic in this kind of population. Two studies have examined the effect of sedative hypnotics with the aim of improving sleep quality during the initial experience of CPAP, with early use predictive of subsequent adherence.(360, 361) Adherence at one month did not significantly improve in a study administering zolpidem for 14 days compared to either a placebo or no treatment intervention,(360) but this study was likely underpowered. In contrast, the use of eszopliclone during the first 14 days was associated with higher adherence and less discontinuation rates across the 6 months study period.(361) However both studies did not actively recruit individuals with co-morbid insomnia.

In addition to these hypnotic approaches, one working group has investigated behavioural approaches in a co-morbid sample that voiced resistance towards the CPAP titration.(362) A retrospective chart review was conducted of 39 OSA patients with co-morbid insomnia, who received a brief daytime exposure to CPAP prior to titration (60-120 min) that aimed to target issues particularly prominent in the insomnia population (e.g. heightened arousal and anxiety, increased wakefulness). The intervention included sleep deprivation (increasing homeostatic drive), imagery and desensitisation and revealed a greater percentage of regular users (≥4hrs on 70% of the nights) compared to controls (59 vs. 20%) Unfortunately, the comparison was made against historical controls (patients before the introduction of the intervention into routine clinical care) rather than a concurrent placebo control group. Again, the intervention was not aimed at improving insomnia, but targeted issues that might be problematic in this population. In a recently accepted study(363) patients who were using a sedative hypnotic for their posttraumatic stress disorder and/or insomnia had higher adherence rates that those who were not. Undoubtedly, there are other issues in this posttraumatic stress disorder group,

however this study adds to the information above that treating insomnia might have beneficial effects on CPAP adherence and yields the importance of a behavioural sleep medicine aspect this area.(364, 365)

1.3.4.2 Interventions to improve CBT-I adherence

Currently there are no studies that have actively set out to design and test an intervention to improve CBT-I adherence. A clearer understanding of what constitutes optimal adherence and how to best measure it is most likely warranted before such studies can be undertaken. Two studies however do lay the path for possible avenues to pursue. One mixed method analysis of 18 individuals completing a sleep restriction programme revealed how the completion of SRT can induce certain side effects, such as sleepiness, fatigue, reduced energy/motivation and even migraines/headaches. Qualitative data corroborated these findings and exposed how individuals struggled with the negative daytime effects induced by their restricted time in bed and how this impacted on their ability to adhere to the programme.(139). Counteracting these effects during the day might lead to improvements in adherence. In Perlis et al. reported increased adherence in the trial arm that received modafinil concomitant with CBT-I, arguably because the modafinil counteracted the experienced sleepiness during the day and individuals were able to stay up till their given threshold time.(218) Adherence was only measured for sleep restriction therapy, however the authors note the use of modafinil might equally be beneficial for the potential side effects experienced with repeated removal from the bedroom with extended periods of wakefulness (stimulus control).

These side effects might be occurring due to reduced initial total sleep time in these individuals in the early stages of treatment. Matthew et al.'s study(217) reported that those who were implementing the treatment were also those who rated their sleep quality poorly in the first week. Thus, the use of pharmacological aids to produce immediate benefits in consolidating sleep might contribute to minimising the impact sleep restriction and stimulus control have on the individual. Using a multiple baseline design, Valieres and colleagues(233) examined the effectiveness of three different treatment combinations of behavioural and pharmacological options: 1) Medication- arm first, then combined medication and CBT-I; 2)Combined treatment then CBT-I alone and lastly 3) CBT-I alone. The presence/absence of 'adherent behaviour' obtained

from the sleep diary, was assessed against seven criteria (e.g. going to bed within 30 minutes of threshold time as described by Bouchard et al. 2003(231)) They report that the average weekly adherence percentages to behavioural strategies (100%= total of 49 possible points achieved) in the medication→ combined group varied between 68.3% to 92.5%. In the CBT-I alone group, adherence varied between 50-85.7%. They describe further that average weekly adherence percentages to behavioural components for the combined→CBT-I group reduced from 92.3% to 72.5%. Although the authors did not make statistical comparisons in adherence rates across treatment modalities, it seems that adherence is largest in the arms combining medication and CBT-I. This might be because of beneficial short term effects to sleep with medication, making adherence to the behavioural components easier. Again this assumption is speculative, because test of differences were not made and results are presented ambiguously.

In summary, initial steps need to be completed prior to the development and implementation of interventions to improve CBT-I adherence. Initial studies do however provide some prospective for intervening when CBT-I is most difficult: i.e. when implementation leads to unwanted side effects during the day.

1.4 Summary

In conclusion, there is a substantial amount of evidence from which to draw preliminary conclusions about potential variables that influence CPAP use, nevertheless, no single variable has been consistently linked to adherence and complex psychosocial interventions are still very much under-researched. In comparison, the literature on adherence to CBT-I must benefit from studies that can establish dose-response relationships and 'optimal' cut-offs, until this is provided, adherence research in this area is unstandardised and will not provide clarity for optimal patient care. It seems then that both areas are virtually at the cusp of an agenda change; CBT-I in terms of standardising the approach to measuring adherence (much like the CPAP literature has attempted with Kribbs's cut off of 4hrs per night on 70% of nights), and CPAP in terms of a more philosophical changes as clearly the approach currently adopted is two-fold: biomedical or psychosocial. This latter point needs to be addressed and could

potentially inform the CBT-I field; and will now be discussed in more detail in chapter 2.

Chapter 2- Integrating Psychology and Medicine In CPAP Adherence- New Concepts?

2.1 Abstract

To date, Continuous Positive Airway Pressure (CPAP) is the most effective intervention in the treatment of obstructive sleep apnoea, but adherence to this treatment is often less than optimal. A variety of factors and interventions that influence and improve CPAP use have been examined. There is increasing recognition of the multifaceted nature of CPAP adherence: the patient's psychological profile and social environment have been recognised, in addition to the more extensively researched patient's treatment and physiological profile. Understanding how these multiple factors impact on CPAP use in an *integrative* fashion, might provide us with a useful holistic model of CPAP adherence. This concept of integration - a biopsychosocial (BPS) approach to health and illness- has previously been described to understand care provision for various chronic health disorders. This chapter proposes an adherence framework, whereby variables integrally affect CPAP use. The BPS model has been on the agenda for nearly 35 years; the presence of poor CPAP adherence was acknowledged in the early 90s- it is timely to incorporate this approach into our care pathway of CPAP users.

2.2 Introduction

'The whole is more than the sum of its parts'

--Metaphysica, Aristotle (384 BC – 322 BC)

Aristotle understood that a holistic approach is crucial to a complete understanding of all aspects of life. More than 2000 years later medicine has seemingly caught up with the value of integration as opposed to reductionism: In the quest to deliver optimal patient care, psychiatrist George Engel's seminal paper published in 1977(366) was a call towards a biopsychosocial model (from here on referred to as the BPS model), recognising that multiple domains (biomedical, psychological and social) integrally and interactively influence health and illness. The aetiology and progress of a disease is influenced by biological processes, psychological experience and social behaviours, which are reciprocally related.(367) To warrant optimal treatment outcome for these diseases, there is then a need for interventions to target all three domains. Haynes stated that *"[e]ffective ways to help people follow medical treatments could have far larger effects on health than any treatment itself."(368, p. 10)* and this importance of adherence has recently been acknowledged by the World Health Organisation (WHO) in 2003.(168) Implicit within these claims, is the need to study adherence within an integrative framework.

Non-adherence is now considered a public health concern: around 30,000 articles have been published on patient adherence, a figure similar to asthma (50,000) and 1/3 of what has been published on diabetes (MEDLINE search with MeSH Headings 'patient compliance', 'asthma' and 'diabetes'). A recent meta-analysis reported that the average non-adherence rate of approx. 25% for complex treatment interventions translates to 188.3 million medical visits in the US resulting in patients failing to implement recommendations and equating to a monetary waste of potentially US$ 300 billion a year.(169) Apart from this economic cost, non-adherence inhibits the evaluation of treatment efficacy at the research level, and can hinder treatment effectiveness at the clinical level.

There has been a major growth in the medical literature related to CPAP adherence following the initial reports on covert electronic surveillance of CPAP use by Kribbs et al.(181) This work is most frequently cited for the commonly used (arbitrary) cut-off of ≥4hrs per night on 70% of the nights. Figure 3 depicts, based on a MEDLINE search, the 362 articles published since 1984 with the terms *'continuous positive airway pressure'* and '*adherence/compliance'* appearing in the title or as MeSH headings (not exploded). A clear surge of articles is evident after Engleman's review in 2003, possibly denoting the recognition that psychosocial variables influence CPAP use.

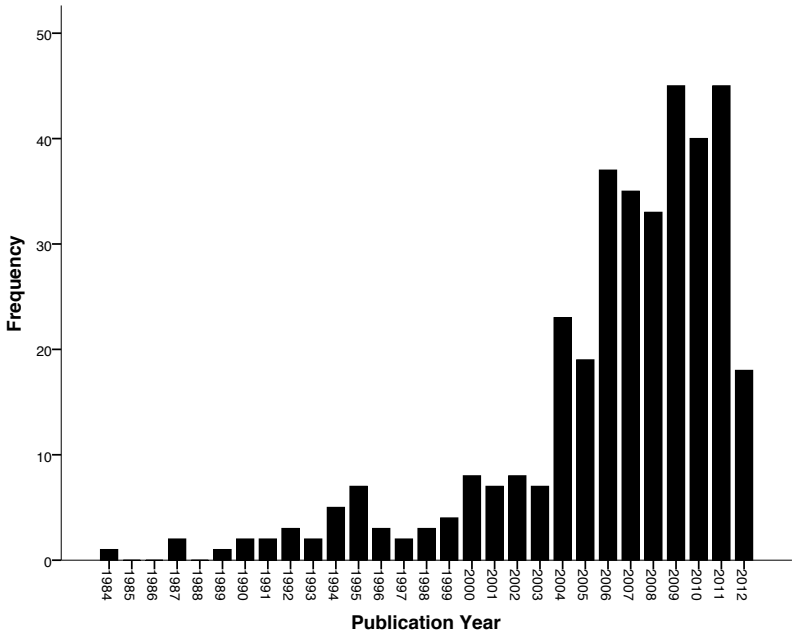


Figure 3: Published articles on CPAP adherence

Our understanding of what mediates CPAP use has increased considerably in the last decade. Undoubtedly, the recognition that various biomedical, psychological and social variables may impact adherence has advanced the field and contributed to the development of more comprehensive interventions to improve use. Despite the awareness of its multifaceted nature, an integrative model has not been proposed for CPAP adherence. As previously proclaimed by Aristotle, a holistic approach requires more than simply taking account of and summing the variables. The objective of this chapter then, is to outline new concepts for the field in light of the current knowledge of CPAP adherence. It is timely to pursue a holistic approach to this problem and consider potential interventions to improve adherence in terms of a biopsychosocial framework.

2.3 A brief review of CPAP adherence

Failure to adhere to CPAP, which is a demanding and invasive treatment, may compromise service delivery and treatment effectiveness. There is evidence of improved treatment effectiveness when the CPAP mask is worn for a large proportion of sleep time.(57) This means that cut-offs of "good adherence" may be suboptimal. However, approximately 25% of patients do not take up CPAP or discontinue in the first 2 weeks after their titration study.(188-192)

Furthermore, for those who continue long term (>1month), 'adequate' adherence (often defined as ≥4hrs on 70% of the nights(181)) can vary between 46-89% depending on definitions of adherence and length of follow-up.(181, 184, 188, 193-202)

Chapter 1 summarised the variables that have been investigated as predictors of CPAP use. Historically, early studies concentrated almost exclusively on biomedical (biological, demographic, treatment specific) variables, such as sleep apnoea severity, age, gender, sleepiness, required pressure.(369) It is evident that apart from severity and symptomatic improvement, most biomedical variables alone have demonstrated little predictive power. Subsequently, studies diverged exploring the role of either psychosocial variables or basic biomechanical factors. To varying degrees, psychosocial variables (such as self-efficacy, illness and treatment beliefs or social support) have been found to predict CPAP use.(369-371) In contrast, the value of biomechanical variables such as mask changes, humidifiers, or machine type have resulted in no or marginal improvements in adherence. Three recent meta-analyses(177, 245, 246) of technological interventions investigated pressure delivery modifications (i.e. autoPAP, BiPAP, and flexible pressure delivery (C-Flex)) yet found no evidence for increased adherence in any modality except for a small, but significant difference of 11-13 minutes in favour of auto-adjusting PAP in all(246) or only cross-over studies.(177) The authors however note that there is little evidence for the use of autoPAP in unselected patients. Additionally, only about 20% of the reviewed papers were clearly described as double blind studies.(177)

Heated humidification can ameliorate adverse upper airway side effects in some patients but there are mixed findings regarding the effect on CPAP adherence(177) and most benefit is found in the first few weeks.(372) The greatest effect of any interventions tested in a randomised trial thus far have

been for psychological interventions such as cognitive behavioural(351, 352) or motivational enhancement(353-355) therapy, with large effect sizes at 1 month (where reported) of d=0.5-1.1, whereas effect sizes for support or educational interventions generally fall within the small to medium range [~d=0.4] or are not significantly different from the comparison group.(242, 288, 369) Although some interventions have been associated with improvements in CPAP adherence, in comparison to a control condition;(288) reported average CPAP usage rarely exceeds 6hrs per night in most intervention studies. However, it is understood from recent dose-response observations that the majority of individuals achieve normal values of daytime functioning at 7.5hrs/night of CPAP use.(57) Of course, even the best options available may be sub-optimal, because they focus primarily on one domain, rather than all domains *and* their interactions. It seems then that the healthcare approach to CPAP use might benefit from a new holistic concept of both the patient and his/her adherence behaviour.

2.4 Conceptualisation of adherence

The transition from the study and reporting of biomedical variables, to psychosocial variables at the research level, parallels a transition in emphasis from compliance to adherence on the conceptual level (see Table 4); a shift in thinking urged by the World Health Organisation (WHO). 'Compliance', defined as *'the extent to which a person's behaviour (in terms of taking medications, following diets or executing lifestyle changes) coincides with medical or health advice'(170, pp. 2-3)* was replaced with the notion of 'adherence', described by WHO as *"the extent to which a person's behaviour […] corresponds with agreed recommendations from a health care provider"*(168, p.18) (see chapter 1 and Table 4). The conceptualisation of an individual as compliant/non compliant overlaps with a stringent biomedical model,(369) which frames the patient's behaviour as somewhat passive, coinciding/not coinciding with the physician's recommendation and influenced by relatively static factors such as age, gender or disease characteristics. The biopsychosocial model, however regards the patient's behaviour as the consequence of an interaction between such static traits and more fluid variables such as beliefs, attitudes, motivation or social circumstances. WHO advocates a more patient-centred approach to adherence; conceptualising the individual's behaviour as a rational (or sometimes irrational) decision, influenced by the interaction between biomedical and psychosocial

variables; in turn, this behaviour may or may not correspond with the agreed recommendations. WHO specifies that '*non-adherence is a multi-determined problem caused by the interplay of four factors. The factors have reciprocal influence on the process of care for chronic conditions'.(225, p. 8)* This approach is akin to a biopsychosocial model and the four factors, the health care team/system, condition/illness related factors, characteristics of therapy and patient-related factors, largely map onto the three domains of the BPS model.

CPAP, then is a complex treatment for a chronic disorder and according to WHO, such conditions are best managed with a biopsychosocial model. Thus far the CPAP literature has largely considered these elements independently rather than as part of an integral whole. The next part of this paper therefore proposes a working model for a biopsychosocial model of CPAP adherence.

2.5 A Biopsychosocial model of CPAP use

2.5.1 Development of the model

Before considering potential interactions, it is important to take account of all possible variables. Figure 4 therefore provides a visual representation of the proposed biopsychosocial model for CPAP adherence.

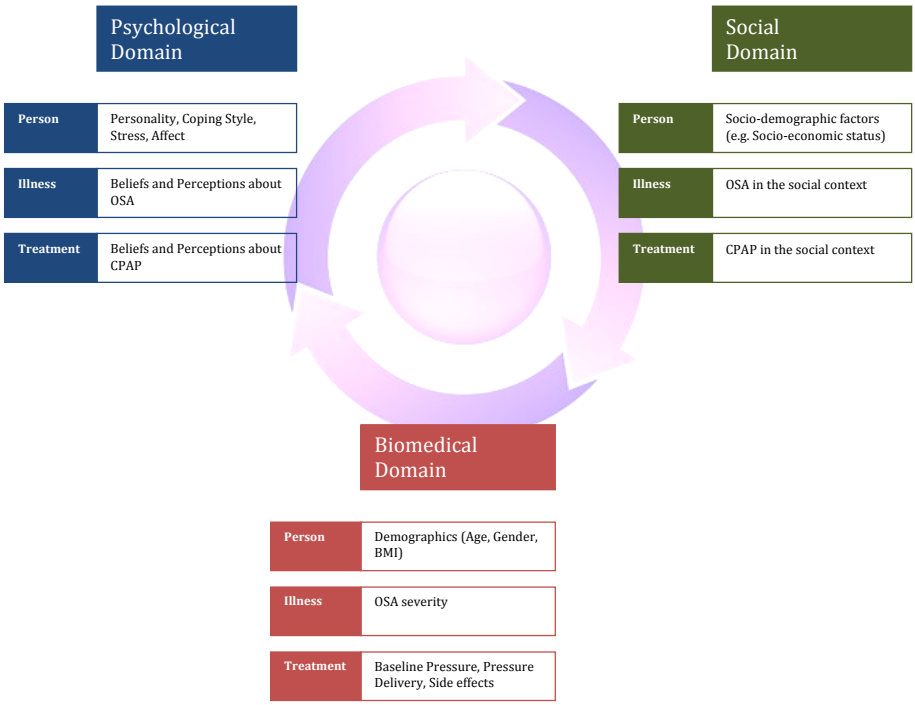


Figure 4: A BPS model of CPAP adherence

As can be seen, the model comprises biomedical, psychological and social domains, each subsuming a person- illness- and treatment- specific level. The biomedical domain encompasses static clinical/medical variables about the patient [e.g. age, gender, BMI, race/ethnicity], the illness [e.g. OSA severity] and its treatment [e.g. pressure delivery, side effects]. The psychological domain on the other hand is made up of variables that are more dynamic and malleable. Coping style, stress and affect, for example are personal level variables that may impact adherence, whereas beliefs and attitudes about their sleep problem [illness level] and CPAP [treatment level], will also inform their adherence behaviour. Such factors may be targeted with psychological interventions. Finally, any individual presents with particular socio-demographic features [personal level]; individuals with different socio-demographic backgrounds might present with different adherence patterns. CPAP does not only concern the individual, but also in the social network around the patient. Thus, adherence might also be influenced by how the illness and treatment is placed into this social framework, how the illness/treatment impacts on their social system and what type of acceptance and support is available from their surroundings.

2.5.2 Integration of the model

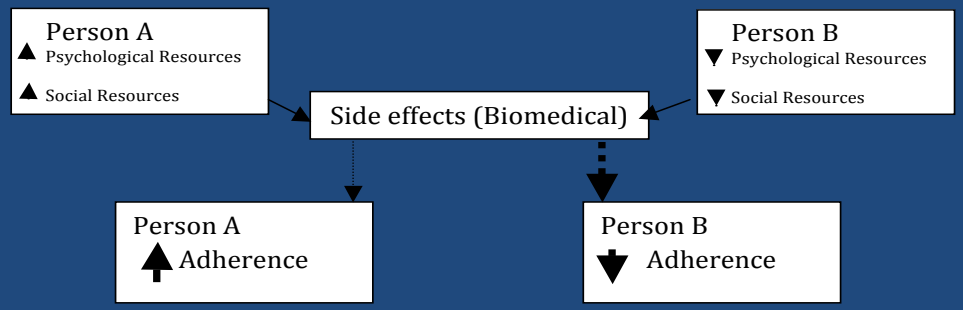
It may be helpful to consider case examples to understand the potential advantages of the model. Here the focus is on the important clinical issue of side effects associated with CPAP use. As highlighted in Box 1 Person A's psychological and social resources make it easier to deal with the side effects he experiences, and so biomedical factors alone explain little of the variance within his CPAP use [weakly weighted dashed arrow in Box 1]. Conversely, Person B has identical biomedical characteristics (side effects, AHI, CPAP settings), but poorer psychological and social resources. Consequently, her side effects have a far greater impact on CPAP use. This individual appears less equipped to deal with the barriers compared to person A. In this case, the side effects (biomedical variable) alone explain considerably more of the variance in CPAP use [strongly weighted dashed arrow in Box 1].

Box 1: Case examples

BOX 1:

Person A is a 45-year-old male, with an AHI of 39 and has recently been provided with a CPAP machine. Throughout the first weeks of use, he experiences residual side effects [not ameliorated within the short term follow-up] such as dry throat, blocked nose and increased nocturnal awakenings. Person A could be described as an individual who takes any situation head on; as a result of this active coping style he informed himself extensively about the sleep disorder and CPAP in internet forums prior to the titration sleep study. He is also feels encouraged to actively seek help with his side effects, whether these are further changes to the mask or any strategies that might help with the nightly awakenings. His wife, who also initiated the first consultation with the GP regarding his snoring, accompanies him to all meetings at the respiratory centre. She supports him emotionally, but also practically where she can- remembering follow-up appointments, maintenance of the machine/mask and suggesting strategies to deal with the side effects. Overall, as a result of his persevering nature and support from his wife, Person A uses the machine throughout the night despite the side effects.

Person B is a 34-year-old female, with an AHI of 40 and has recently been provided with a CPAP machine. Throughout the first weeks of use, she experiences side effects [not ameliorated within the short term follow-up] such as dry throat, blocked nose and increased nocturnal awakenings. Person B could be described as an individual who tends to shy away from difficult situations. She has found the diagnosis of OSA and prescribed CPAP treatment a change difficult to cope with. She tries not to ask for help, as she feels this may be a sign of weakness. She is currently single and concerned with the potential negative impact CPAP might have on a new relationship. Her social support network is confined to her mother, who has limited understanding for her daughter's situation, blaming her for neglecting her health. These concerns also further fuel her nocturnal awakenings initially caused by the CPAP machine. She tries to use the machine for the first few hours, but generally gives up when she wakes up at night frustrated and feeling lonely.



The examples also highlight the reciprocal interaction between biomedical, psychological and social factors: For person B the side effects might not only impact on adherence, but might also have a reciprocal effect on her motivation/feelings of isolation, leaving her even less equipped to deal with the side effects. Additionally, the nightly rumination about these concerns might further increase the wakefulness during the night, possibly potentiating the impact the side effects have during the night. The result: an interacting cycle of effects across domains of the biopsychosocial model of CPAP adherence.

As far as treatment is concerned, then, one can conclude that it is likely to be less than optimal to consider biomedical, psychological or social variables in isolation. Support requirements for person A might be limited to technological interventions to eliminate side effects as much as possible, whereas support for person B might include resources for improving psychological and social functioning to mitigate the impact of side effects.

2.5.3 Implications of the model for research

There is now need for the investigation of possible interactions amongst variables that have previously been examined in isolation (see chapter 1). The systematic investigation of these interactions will have implications at the research level, and may benefit from more sophisticated statistical methods, for example structural equation modelling(373). This technique enables assessment of causal relationships and interaction effects between variables; and calculates the effects of any latent determinants.(374) Analysis of this kind can handle complex interactions and will permit greater determination of proportions of variance explained by BPS profiles (phenotypes) that might be common to a group of individuals. This task might also be facilitated by the use of cluster analysis. A recent health psychology review described this method as:

'[t]he process of taking a heterogenous sample of entities […] and forming relatively homogenous groups' through the organisation of *'large quantities of multivariate information. Labels can be assigned to the subgroups, making the data more manageable for the individual researcher […]'* This technique then allows for *'identification of groups that might best benefit from interventions or further research.'(375, p.330)*

Clearly, it will be important, both for research and clinical practice, that *all* relevant variables are measurable. With this in mind, it should be noted that there is a sound psychometric tradition of psychological measurement that could be seemingly adapted to this population. Brief scales measuring coping style,(376) self-esteem,(377) personality,(378) relationship intimacy(379) are examples that would compliment scales/measurements that are already well established to identify the biomedical characteristics of the CPAP user. This includes instruments such as the Epworth sleepiness scale (ESS),(5) where individuals rate the chances of falling asleep in eight situations. Although the ESS has been criticised for its moderate correlation with objective measures of sleepiness, like the multiple sleep latency test(380), the message here is that the scale is easily implemented and has face validity, and has been effortlessly integrated into the care pathway by treating physicians.

2.5.4 Implications of the model for clinical practice

To understand the conceptual shift that clinicians will have to make, Figure 5 describes a typical clinical scenario- tackling nasal stuffiness as a possible side effect. The top box, model 1, is an illustration of the multifactorial approach, whereas the lower box, model 2, displays an integrative approach. The clinician taking an integrative approach may form an understanding of the reciprocal effects between biomedical, psychological and social domains. The exploration of these interactions will enable the clinician to answers to question left unanswered in model 1 or at least to provide a pathway for investigation. Do side effects reduce the individuals' motivation, despite motivational support prior to CPAP initiation? Does the increased frustration with the CPAP machine lead to tension between the patient and the partner, resulting in a reduction in partner support? Does this in turn leave the individual feeling isolated and unable to make use of any remaining resilience?

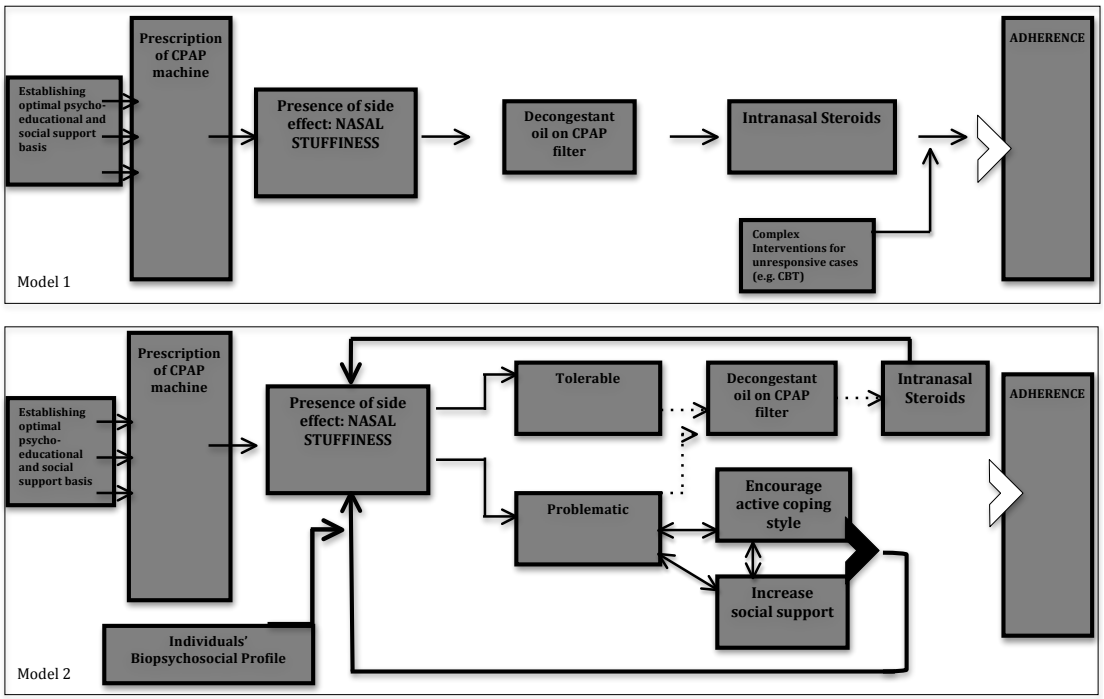


Figure 5: Potential approaches to nasal stuffiness as a side effect.
Model 1 indicates the simple multifactorial approach to CPAP adherence, adapted slightly from Engleman et al 2003.(369) Side effects are eliminated with technological (biomedical) approaches. Model 2 highlights a biopsychosocial, integrative approach where side effects, when problematic and/or unresolved by technological interventions, are tackled by inclusion of psychological (increase personal reliance) and social (increase social support) interventions. This model also indicates the interaction between these domains (two-way and solid arrows) as well as one-way relationships displayed in previous models (dashed arrows). The individual's BPS profile may inform the integration of psychosocial interventions prior to the presence of side effects, rather than after problems occur as described in model 1.

Of course, clinicians may be concerned that highly tailored interventions for each CPAP patient may not be feasible, or affordable. Therefore it is important to recognise that one potential merit of the BPS approach, in contrast to a multifactorial model, will be the development of tailored interventions that can ultimately be applied at the population level.(381, 382) That is, once possible BPS profiles have been conceptualised [at the population level], the clinician may be able to match the patient's profile [individual level] and so determine best care.

2.5.5 Potential resistance to adopting the BPS model

There is anticipated resistance to implementing the BPS model: many will surely argue it is labour, time and cost intensive, however it is important to remind ourselves of the labour, time and cost implications of non-adherence. What is currently done equates to sub-optimal adherence rates. There is currently no estimation of the direct cost of CPAP non-adherence, however a recent review

estimated that an average non-adherence rate of 24.8 % across the selected treatments would, in 2001, have translated to an approximate cost of US$ 300 billion and the adherence rates for treatments of sleep apnoea were the lowest of the reviewed treatments.(169) Additionally there are indirect medical costs of untreated OSA linked to cardiovascular events and mortality,(70, 82) and increased non-medical costs such as automobile accidents or work absenteeism.(383, 384) It is at this time not possible to estimate the exact direct and indirect cost of CPAP non-adherence compared with the cost of the implementation of a BPS approach in the clinical service. However, it should at least be evident that what is currently implemented to improve CPAP use is not sufficient, and that a major paradigm shift is now required.

2.5.6 What has been done so far?

To date, there has been no reference to an integrative model of CPAP adherence; however, there is some evidence in the current literature to be reviewed that provides support for the superiority of examining the interactive effects of variables from different domains.

2.5.6.1 Reviews

A few reviews have previously explored the issue of CPAP adherence and its various dimensions. Engelman and Wild presented a clinical practice and research formulation for managing problematic CPAP adherence.(369) The recurring theme that a reductionist biomedical approach to adherence is not sufficient, was also discussed in this review. The authors state that some biomedical variables correlate weakly with adherence, however, and more importantly, these variables are associated with certain underlying cognitive attributions and cost-benefit ratios e.g., the cost-benefit ratio for CPAP adherence might differ between males and females. There are indications in the text, that the authors of this review advocate a biopsychosocial approach to CPAP adherence:

'The enhanced value of adherence models incorporating cognitive constructs is that variables immutable under a biomedical model, such as AHI or Epworth sleepiness score, become plastic [58]. While the biomedical model reifies these weak determinants as independent objective entities, within a

cognitive model the emphasis is shifted from the numeric value to the patient's perceptions of these values.'(369, p. 92)

The review does not offer a detailed and/or explicit account of how this may occur or even call for a research agenda that incorporates these kinds of investigations. In fact, the point made above, does not feature in Engleman and Wild's practice points or research agenda, and has clearly not translated into the CPAP literature, where the majority still clearly distinguish between and fail to integrate the biomedical and psychosocial domains. Thus by focussing upon the sum of the multifactorial parts of their model, the strength of Engleman and Wild's review relates to those parts. However it does not explicitly present a biopsychosocial, integrative model of these variables. As a consequence of their suggested approach CPAP non-adherence is targeted at the individual level, rather than the population level (see above). Furthermore, they argue that complex, cognitive interventions are 'saved' for difficult or unresponsive cases. In contrast the BPS model outlined here will hopefully contribute to the identification of BPS profiles that can be implemented before individuals encounter problems and possibly prevent them from exiting the system (see Figure 5). Lastly, the model presented here does not discard biomedical variables that are not univariate predictors of CPAP use, but calls for a re-evaluation of these variables within a BPS, rather than a univariate framework. In contrast, by emphasising only those variables that are univariate predictors, Engleman and Wild implicitly discard those that do not. Nevertheless, their review offers preliminary support for a BPS model and hopefully this chapter extends their attempts, by providing a more detailed and explicit account with reference to research studies investigating interactions between all domains.

In a second review, Sawyer and colleagues(242) describe CPAP adherence in the context of age: for example in younger age groups the family context is an added factor (positive or negative) in the complex picture of CPAP adherence and variables such as parental education, attitudes and beliefs can impact on the child's CPAP use, whereas for older adults any potential reductions in use may be mediated by other factors that increase with age, e.g. insomnia. It is thus imperative that interventions to improve CPAP adherence are tailored to allow for age- and development-specific factors. Although not explicitly mentioned in Sawyer et al.'s review, the authors are drawing upon a biopsychosocial framework, by assuming an interaction between age and

psychosocial characteristics. Further exploration of these relationships should provide an understanding of how variables might have a differential and interactive effect on adherence across age groups. For example, younger CPAP users usually have family members/caregivers that can offer support in the home, whereas older adults are likely to be living alone. For the older age group, there might be a different meaning attached to using a machine to control one's breathing ('getting old'), which may lead to dysfunctional beliefs and attitudes about the disorder and treatment, whereas the younger age group might consider wearing a CPAP machine as 'uncool'. With an increase of insomnia symptoms with age, older CPAP users might struggle during the night, and increased frustration and worry might further exacerbate the wakefulness. This in turn may disrupt the partner's sleep and potentially reduce the effective support they are able to provide. These factors are still very much under researched; nevertheless it is clear the psychosocial profile of individuals in each age group might vary and adopting a BPS approach could provide insight into what factors may be important across these groups.

2.5.6.2 Research studies

A few research studies have been published with reference (albeit implicit) to an integrative model of CPAP adherence. Using an interesting day-to-day study design, a recent study asked individuals to complete measures of spousal involvement (pressure, support, and collaboration), and examined the daily relationship between ratings of spousal involvement and subjective adherence.(338) The relationship between adherence and support was moderated by disease severity (AHI): support was only predictive of next day adherence in those within the highest quartile of AHI scores. The authors conclude that perhaps those with lower disease severity experience less daytime symptom improvements and any support from the spouse is considered unhelpful and might not merit attention from patients. Using the day-to-day analysis, the authors were also able to investigate the directional relationship between adherence and involvement. Interestingly, spousal collaboration was increased after nights with poor adherence, and the authors relate this to possible sleep disruption caused by emerging of nocturnal symptoms (e.g. snoring) with non-CPAP use. Although the caveat of this study is the use of subjective daily

adherence, this study highlights the possible intricate relationships between spousal involvement, disease severity and adherence.

A further example of a BPS approach to CPAP can be found in a paper published by Olsen and colleagues(301), who expanded on previous attempts to understand the relationship between certain health beliefs and CPAP adherence. The first study(308) found variables from the Health Belief Model (HBM) measured only 1 day after titration were good predictors of CPAP use at 1 month. This social cognitive model describes the likelihood of the individual's behaviour is influenced by representations individuals have of their illness (in terms of severity and their own susceptibility) their treatment (e.g. benefits and barriers of engaging in it) and their self (e.g. perceived control). Olsen et al. found a similar relationship when the variables of the model were measured prior to CPAP initiation. They reported that patients' outcome expectancies and risk perception were independent predictors of CPAP use. The HBM variables alone, explained 21.8 % of the variance. Reflective of previous findings,(308) biomedical variables alone were not predictive of adherence. However, when these were included in the model alongside the HBM factors, the explanatory power rose to 31.8%. Although biomedical variables were included in the original Health Belief Model, they have not always been acknowledged. Rosenstock, the author of the original HBM article, stressed the importance of considering the interactive relationship between health beliefs and demographics and that certain cognitive attributions might be differentially distributed across these demographic subgroups.(385)

Other studies have shown when both biomedical and psychological variables are included into the same statistical model - albeit sometimes without a theoretical underpinning - more variance is explained.(286, 287, 300, 304, 333) Interestingly, Poulet and colleagues(300) using a decision tree analysis- by nature a method to investigate interactions- were able to correctly predict 86% of the non-adherent patients with three baseline variables: emotional reactions, age, and maladaptive beliefs. Younger individuals reporting less emotional reaction to symptoms and with less accurate beliefs were correctly identified as non-adherent to 85.7% certainty. However in the same individuals with more adaptive beliefs, the accuracy rates for predicting adherence and non-adherent individuals was almost identical (58% vs. 42% respectively). Unfortunately, these accuracy ratings at the last node of the decision tree were based on a small

number of individuals (i.e., less than 20 individuals from the initial sample of 122 scored were available in each node of the last stages of the decision tree). However, this study does suggest the usefulness of examining variables from both the biomedical (age) and psychological (beliefs) domain. The authors conclude that this type of analysis might be useful in informing individually tailored interventions.

Lastly, Means and colleagues examined the relationship between ethnicity and adherence in a sample of 501 military veterans with and without a comorbid mental health disorder.(290) African Americans (AA) had lower CPAP use (approx. 1hr) at both one and three months post treatment initiation than the Caucasian group (CA). Interestingly, African Americans with a mental health disorder had a significantly lower adherence rate than all other groups (AA without a mental health disorder and all CAs), indicating an interaction effect of ethnicity and presence of a mental health disorder on CPAP adherence.

These examples illustrate how certain groups of CPAP users might present with specific psychological and social attributions. Until now, this review has- mainly for simplicity reasons- considered the variables uniquely nested within each domain (e.g., AHI is a biomedical variable and not considered within the psychological or social domain), however it is important to remember that some biomedical variables may overlap with the psychological domain,(369) e.g., side effects and symptomatic improvement. That is, these biomedical factors are malleable to psychological (placebo) effects and two recent studies provide preliminary evidence for this.(1, 251) A recent patient-level meta-analysis of three randomised placebo-controlled crossover trials indicated that the effect of adherence on improvement in daytime sleepiness might be attributed to both a real effect and expectation of benefit. Higher use of CPAP was associated with improvements compared to low CPAP users, interestingly though a similar, but less extreme pattern was found in the placebo arm, indicating that some of the symptomatic improvement might be a result of a placebo effect.(1)

Likewise, individuals might be more likely to report side effects when warned of these in the education session, the ‘nocebo effect’ and this effect might be increased by certain personality traits.(386) One study has investigated the relationship between personality type and CPAP side effects (e.g. blocked nose, dry throat etc.) and their impact on machine use.(251) In this cross sectional study 247 OSA patients, who had been using CPAP machine for longer

than 6 months, were assessed for Personality Type D (described as a tendency to experience negative emotions coupled with inhibiting the disclosure of these emotions to important others), the experience of side effects in terms of frequency, magnitude and perceived impact on adherence, as well as objective machine use. Individuals were then separated into those with and without personality Type D. Those with Type D were more likely to report certain side effects, judged them as more severe and also indicated the impact on adherence was greater than those without Type D. Although there was a significant difference in objective use per night (4.8 hrs Type D vs. 6.3 hrs non-Type D, $p<0.001$), no information was given about the interaction between side effects and personality type and their influence on objective hours of CPAP usage. An additional limitation is the cross-sectional rather than prospective nature and it remains difficult to establish whether personality type D was perhaps the result of non-adherence (i.e. sleep deprivation).

In summary, a BPS model of CPAP adherence needs to account for not only the multitude of biomedical and psychosocial variables, but also clarify how they interact. This would result in a holistic model describing certain individuals with a specific biopsychosocial profile and which groups might benefit from specific tailored interventions based on these interactions. This might be a challenge- a challenge to implement as well as a challenge to convince sleep professionals, however if the impact of CPAP non-adherence is considered, it is a challenge that seems very worthwhile. With this in mind, it may be helpful to highlight how other health services have benefitted from this shift in perspective towards a biopsychosocial approach.

2.6 The success of the biopsychosocial model- its application in health and illness

The biopsychosocial model, introduced to the medical domain by Engel, was envisaged as an addition to the medical model, rather than its replacement. However, the viewpoint that health and illness are defined by biological processes, resulting from injury, biochemical imbalances, bacterial or viral infections(366, 387) often overlooked important individual experiential factors(388) and/or significant social or cultural variables,(389) which merit consideration alongside biomedical factors. The adoption of a more integrative

framework has facilitated health improvements elsewhere, e.g., peptic ulcers(390, 391), antisocial behaviour disorder(392) or chronic pain.(393, 394) Chronic pain and peptic ulcers have been selected here as illustrations.

A peptic ulcer is an ulcerated area within the gastrointestinal tract (stomach, duodenum, or oesophagus). The disease exemplifies the biopsychosocial problem, initially believed to be a psychosomatic disorder, caused predominately by stress and personality,(395) yet with the discovery of the bacteria Helicobacter pylori (H. pylori) as the cause of peptic ulcers(396), it was then defined as being purely physiological. One reductionist viewpoint had been replaced by its mirror image.(397) Much later came the recognition of the interplay between these two viewpoints and that H. pylori is one piece in the puzzle of a biopsychosocial framework.(391) In fact a recent well designed animal model study(398) examined the synergistic relationship of H.pylori and psychological stress, by comparing the inflammation in stomach tissue of mice between 5 conditions (control, bacterial infection and psychological stress alone and in combination as well as psychological stress + treatment). Interestingly, psychological stress alone was not statistically different from the control condition, but it had additive effects in the combined condition, which is suggestive of a possible interaction effect of H.pylori and psychological stress on the development of peptic ulcers.

Chronic pain, in contrast to peptic ulcers, was first considered as a purely physical phenomenon. The early Cartesian model offered an explanation of pain perception as a biomedical, physiological process, whereby the experience of pain occurs as the nociceptive signal travels from the point of injury via the spinal cord to the brain. The majority of theories left little room for psychological and/or social factors, unable to explain phenomena such as the varying relationship between injury and pain, phantom limb pain, and persistent pain after tissue healing.(393) With the development of the gate-control theory(399, 400) and its successor the neuromatrix theory(400) it was understood that pain perception is influenced by top-down processes from the brain. The neuromatrix is the connection of different areas of the brain: cognitive-evaluative (past experiences, cultural learning, anxiety, attention), sensory-discriminative (sensory input) and motivational-affective (hypothalamic-pituitary-adrenal system, immune system) which, when activated, result in pain perception. This integration of multiple domains has made it possible to explain

phenomena such as phantom limb or chronic pain as '*the result of neural mechanisms gone awry*'.(401, p. 1678) Today it is understood that psychosocial variables such as cognitions, emotions, social support in addition to sensory input (nociception) can and do influence pain.(402, 403) A recent review in fact highlighted the mind-body interactions in pain with reference to the psychological concept of catastrophising. Interestingly, they describe how catastrophising is associated with neuro-endocrine, neuro-immune, psychophysiologic, and functional neuro-anatomic changes in areas that are directly related to sensory pain perception.(404)

A reductionist view of the aetiology of disease is limiting, whereas integrating biomedical and psychosocial perspective has led to a more holistic understanding of both PU and chronic pain. These examples provide a description of the aetiology of an illness, yet to fully understand the merit of adopting BPS framework for CPAP adherence, it is necessary to consider the implementation of such a model for adherence in general.

Engel claimed that all three BPS domains must be taken into account in "every health care task".(405, p.179) Management of adherence is a health care task, acknowledged by WHO as more important than any other medical treatment for improving patient outcome. Christiansen and colleagues(406-410) have utilised an integrative approach for the understanding of adherence to treatments for end stage renal failure patients. Their patient-context theory states that patient characteristics alone are not associated with adherence and it is the interaction between these characteristics and the treatment context that is most useful in understanding adherence to haemodialysis. This treatment takes many forms and varies in the degree of behavioural control required: centre haemodialysis, where the patient takes on a relatively passive role, home haemodialysis, where the patient is required to participate in some of the decision making, and lastly continuous ambulatory peritoneal dialysis, (CAPD) which is the most behaviourally intense in terms of input needed by the individual (e.g., bag changes 4 times a day). Congruence between the patient's preference for involvement and type of treatment is more predictive of adherence to treatment than type of dialysis alone. Patients who favoured a more active coping style(409) or were more vigilant with information regarding their health,(406, 408) were adherent to dietary and fluid intake requirements in the treatment context requiring a more active role (e.g. home or CAPD) than

in the more passive context (centre haemodialysis). The opposite pattern was found for those preferring a more passive role within their treatment. The interaction between patient preference and treatment context is crucial for adequate adherence.

This example highlights how a reductionist perspective, where adherence is considered within the treatment context *without* taking the individual's preference into account, is less beneficial. This would be equivalent to trying to make sense of pain perception by considering sensory input only, or understanding peptic ulcers through H.pylori infections alone.

2.7 Summary

This review has provided an overview of how a biopsychosocial model could be adopted for the management of CPAP adherence on both a research and a clinical level. Following the success of this approach in other disease entities, both in advancing knowledge and in improving clinical practice, this chapter calls for a biopsychosocial approach to CPAP adherence and perhaps to OSA management more generally. CPAP adherence is multifaceted, and that patient-centred research is vital to advance the field, there is now a need for a paradigm shift towards a holistic approach, where consideration of the interaction of biomedical, psychological and social factors is the clinical norm.

2.8 Aims of the thesis

In the context of this narrative literature review three core aims can be identified to enable the progression of our understanding of CPAP adherence. Firstly, there is a need for identification of the largely under-researched psychological and social variables. Secondly, research needs to focus on not only the multi-dimensional, but also the holistic nature of CPAP adherence, understanding the possible interactions between variables of all domains of the BPS model. Lastly, there is a need for developing brief, reliable and valid scales to facilitate the identification of BPS profiles within the clinical setting. The dearth of information on adherence to CBT-I outlined in the introductory chapter highlights how these issues can easily and should be extended to the insomnia field. These conclusions provide the basis for the three overarching thesis aims.

Aim 1: The first aim is to identify further psychological and social variables predictive of adherence to both CPAP and CBT-I. A unique opportunity is provided by in depth exploration of the patient's experience of implementing these treatments. The two qualitative studies outlined in chapter 3 and 4, will provide greater insight into the psychosocial aspects, real life experiences of the treatment of both insomnia and OSA, and direction for further exploration of interactions between biomedical and these psychological and social factors.

Aim 2: The next step is concerned with discovering the full potential of the BPS model by considering possible interactions between biomedical and psychosocial variables. As described above and in the introduction, the literature has reported on the relationship between adherence and outcome; specifically revealing that increased adherence may lead to improvements in daytime sleepiness. In addition to a physiological basis for this relationship, psychological effects might play a role, i.e. the relationship is influenced by an expectation of benefit. The identification of both these physiological and psychological effects on the dose-response relationship between adherence and outcome was the main aim of chapter 5 as a published paper. Three placebo-controlled trials were combined to quantify these effects. Most notably, finding improved outcome with increased use of both CPAP and placebo would indicate the presence of physical as well as psychological effects based on an expectation of benefit.

However, as noted above, the relationship between adherence and outcome is likely to be bidirectional; that is, increased use can lead to increased improvements, and improvements can also spur on adherence. This has implications for the aim of chapter 5 within the context of this thesis. If the results of chapter 5 show that sleepiness outcome is affected by an expectation of benefit over and above the physiological effects, then sleepiness as a predictor of adherence should not only be considered within the biomedical domain, but also as a psychological factor, supporting the need for the implementation of the BPS model. Furthermore, and keeping in mind that there is potentially a bidirectional relationship, if both physical and psychological effects affect how increased adherence leads to improvements in sleepiness, then ultimately both physical and psychological effects affect how improvements in sleepiness also lead to increased adherence. Thus, the aim of this chapter is to identify the psychological and physical effects on the relationship between

adherence and outcome, to provide evidence for the BPS model of CPAP adherence.

Aim 3: The last aim considers the seamless translation of a BPS approach to clinical care. The development of brief, reliable and valid tools to identify psychological and social variables to be added to the already established biomedical measures in routine clinical care will facilitate this task. Chapter 6 will describe this process for a scale assessing components of the transtheoretical model (stage of change, self-efficacy, decisional balance and processes of change). The insomnia population will provide the context for this last aim, however efforts will be made to develop a scale that is easily transferable to the CPAP adherence field and potentially other health domains, thus providing a holistic approach not only to adherence CPAP and CBT-I, but to adherence in general.

The next sections will be concerned with the evaluations of these three aims in the context of four experimental chapters. The aims and conclusions that are relevant to each chapter in isolation will be discussed within each section. How these results relate to these core thesis aims will be evaluated in the overarching discussion in chapter 7.

Chapter 3- *"It's logical, it's rational, and yet I don't want to do it"*- A longitudinal qualitative analysis of the experience of using Continuous Positive Airway Pressure

3.1 Abstract

This study set out to examine the experience of patient adherence following a diagnosis of OSA. Interviews were conducted with 11 participants, who had recently been prescribed a CPAP machine and were attending the sleep clinic for their titration study. Six individuals completed three interviews (pre-titration, 1 week and 3 months after CPAP was introduced into the home). Topics covered in the interviews included: the experience of having sleep apnoea and what enabled them to use their CPAP machine. Audio files of the interview were transcribed verbatim and evaluated using thematic analysis. Three key themes emerged in the interviews: 'internal conflict around acceptance and adherence', 'integration of CPAP into life' and 'motivators and resources for CPAP use'. It is apparent from this study and previous qualitative work, that CPAP is a demanding treatment. Perseverance is vital for the integration of CPAP into patients' lives. Consideration of possible interactions amongst these factors may inform service improvement and future research.

3.2 Introduction

In their guidance documents on complex interventions, the Medical Research Council (MRC) alluded to the importance of considering qualitative, as well as quantitative, methodologies for development and evaluation of complex treatments.(411) The qualitative approach has been valuable for undersanding adherence to treatments ranging from medication after renal transplants(412), home exercise for chronic pain(413) and type II diabetes(414) to name a few. A number of studies have investigated the experience CPAP treatment, undoubtelbly a complex intervention, using qualitative approaches. The majority has focused on one single time point, assessing beliefs and attitudes either prior to treatment experience, or after treatment is initiated.

Broström and colleagues interviewed 23 CPAP users who had initiated CPAP between 2 weeks and 182 months prior to the interview.(415) Using a qualitative content analysis approach this study identified certain facilitators and barriers for CPAP use. Wanting to avoid symptoms and health consequences of OSA, fear of disturbing others' sleep, positive attitudes towards treatment; seeing improvements, spousal support and healthcare personnel were aspects that facilitated adherence. Negative attitude towards treatment, practical problems with CPAP, side effects, negative psychological effects of the treatment, insufficient spousal and healthcare team's support created barriers to CPAP use.

Although this study provides important information about factors that influence CPAP use, two limitations were apparent. There was a large variation in the time intervals between CPAP titration and interview across individuals (2 weeks-182 months) adding great variability to the overall results and it is unclear which factors might be more/less important at different stages of treatment. Secondly, the questions defined a priori were strikingly similar to the subthemes. The questions were structured around asking the participant to describe a situation that *facilitated/ created a barrier* for CPAP use at a general level and then in relation to situations involving the *spouse* and *healthcare personnel*. This casts doubts as to whether the emerging themes from were truly patient-generated.

In 2006, Dickerson and Kennedy interviewed 17 CPAP users who had attended a CPAP support group.(416) Using a phenomenological approach (Heideggerian hermeneutics), individuals described group support as beneficial

in facilitating motivation to persist with treatment despite its difficulties, to obtain the benefits CPAP could produce. Secondly, individuals described getting used to the machine and the need to integrate it into their lifestyle; tips for how to do this were provided by the 'expert/long-term' CPAP users. Individuals described how they felt empowered by group support, felt less isolated in dealing with this illness and treatment; and benefitted from listening and telling stories and giving/obtaining practical advice. The support group was defined as a community of CPAP users that could encourage, make people feel less isolated and was a forum for exchanging advice and providing hope. Overall, the authors argued that this format might provide the support and knowledge that could be introduced into routine clinical care. One limitation of the study was that individuals were recruited by advertisements, thus participants might have been biased towards a more open discussion about their difficulties with the machine.

Smith et al. interviewed 21 CPAP users (3-39 months since initiation) and their partners in relation to their learning needs and barriers of use.(417) Content analysis of the interview data revealed that both patients and their care-givers reported improvements in sleep quantity and quality through use of CPAP. Barriers for implementation reported by the caregiver were of a psychosocial nature (impact on routines, travel), whereas patient reported barriers concerning the practical impact of machine use (i.e. side effects). They also asked individuals about the type of information most desired from the health care team. Information about sleep apnoea and its consequences on health was most frequently mentioned, followed by knowledge on implementation of CPAP into everyday life and other more practical issues concerning machine use. However, the questions used were similar to the emerging themes (e.g. open-ended questions about adherence, benefits, problems, current knowledge informational needs) and naturally this type of approach might be biased by demand characteristics.

Veale and colleagues analysed interviews with thirty OSA patients, 15 of whom were currently using CPAP.(418) Discourse analysis using the following three methods: content analysis, propositional (assessing the proximity of words/terms) and lexical analysis, revealed how individuals witnessed improvements in their sleep, despite reporting difficulties using the machine, especially those who were currently working, or travelled frequently. A number of participants considered themselves ‘ill’ as a result of accepting the medical

treatment. Unfortunately CPAP adherence was not the primary outcome of investigation in this study, but rather how the diagnosis of OSA impacted on individual's quality of life.

Van de Mortel et al.(275) used grounded theory to investigate the experience of the titration night and subsequent CPAP use. Interviewing 19 individuals, nine of whom reported not using the machine, the authors deduced two theories from the data: 1) benefit derived from treatment translates to increased use of the CPAP machine and 2) poor experience during the titration night leads to poor CPAP adherence.

Support from the medical team or CPAP suppliers was reported as important in the process of accepting and using CPAP in a number of the qualitative studies, with some patients experiencing levels of support below their expectations.(e.g., 415, 417). Shoukry et al.(419) were interested in the experience of a particular type of medical team: pharmacists. Twenty patients who had been provided with a CPAP machine from the pharmacy –based CPAP service, were interviewed and using phenomenological analysis, the authors described the patient's general experience of from-diagnosis, to the titration study and then CPAP use. This overall CPAP experience was similar to the studies described above, and most patients were generally satisfied with the service provided by the pharmacy. Patients valued the convenient access, and the majority considered the personalisation of care offered by the pharmacist suitable for their needs. Unfortunately patients were interviewed between 0.5 and 84 months of receiving their CPAP machine. Because of such a large variance in time intervals and thus heterogeneous sample, specific conclusions are difficult.

In addition to these cross-sectional investigations, a few studies have embarked on a longitudinal qualitative investigation of CPAP use. Broström et al.(420) presented a longitudinal case study providing in-depth information about the experience of implementing CPAP treatment from a patient and his partner's perspective. Semi-structured interviews were carried out with the two individuals at four timepoints across 6 months (before CPAP , two weeks, three and 6 months after CPAP initiation). Using a phenomenographic approach, this study found that CPAP acceptance and adherence was slightly inhibited by the lack of initial information given at diagnosis. As treatment progressed further emotional and educational support from the CPAP-nurse enabled the couple to

fully understand sleep apnoea and its consequences, overcome negative social reactions (e.g. strain on the relationship) and stressors (e.g. negative health consequences of OSA, side effects of CPAP, fear of being perceived as odd) - which increased CPAP adherence in the long-term. This study provides a fascinating insight into CPAP adherence from a longitudinal and couple's perspective.

Another study in 2007 presented data on 20 newly diagnosed OSA patients followed through from titration to one and three after CPAP initiation(421). Using an interpretive phenomenological approach(Heideggerian hermeneutics) the authors identified five themes. The philosophical foundations of this analytic approach lay within phenomenology: understanding how people make sense of a particular phenomenon; hermeunetics: the theory of interpretation, and ideography: the study of the particular both in the sense of detail and a particular group or context(422) and in this context allow for a more detailed, interpretive account of the individual's experience of using CPAP. Throughout the treatment process, individuals found using the CPAP machine difficult, reporting common side effects such air leakage, marks from CPAP straps and disrupted sleep (theme 1). Pushing through these difficulties required persistence and perseverance on the patient's behalf (theme 2). Individuals seemed challenged by identifying improvements in their symptoms, especially in the early stages of treatment. As they progressed through to the 3-month interview, changes were more apparent, often endorsed by improvements witnessed by family members or non-adherence (theme 3). Problem solving and gaining support, especially from family and CPAP providers, facilitated pushing through these difficulties. (theme 4). Based on a cost-benefit evaluation CPAP was either integrated into their lives or the device was abandoned (theme 5). Lastly the authors describe a constitutive pattern, namely pushing through difficulties by obtaining a positive mind-set. Although this comprehensive study presents a detailed account of the experience of implementing CPAP, the themes are not clearly defined with large overlap in places (e.g. theme 1 "trouble using CPAP" and 2 "needing to persist through initial and recurring frustration"), which hinders the provision of clear directions for treating clinicians and future research. Furthermore, the second interview was conducted 1 month after CPAP initiation; there is evidence to suggest that within the first month, 25 % of individuals stop using CPAP and adherence

patterns are relatively well established after 1 month. Interviewing CPAP users at a shorter time period after the titration (e.g. 1 week after) might provide important information about factors that influence CPAP use.

In addition to Dickerson's study, only one working group has evaluated the experience of CPAP users in a longitudinal design.(423) This study drew on aspects of social cognitive theory as a conceptual framework to further understand what factors differentially contribute to adherence and non-adherence to CPAP. This model describes knowledge, self-efficacy, outcome expectations and facilitators of/barriers to change as strong predictors of behaviour change. Fifteen newly diagnosed OSA patients were interviewed post-diagnosis and after a one-week experience of CPAP use. Interviews were firstly content analysed, identifying themes at the individual level. The sample was then separated into adherers (≥ 6hrs/night CPAP use) and non-adherers (< 6hrs/night CPAP use). Differences between adherers and non-adherers emerged in terms of risk perception, symptom recognition, self-efficacy, facilitators and barriers outcome expectations and treatment goals. Adherers were generally considered OSA to be more aware of the symptoms and potential consequences, and had increased confidence. These individuals were also more likely to set treatment goals and focus on positive outcome expectancies in order to overcome any barriers. The strength of this multi-method study was the blinding of the investigators to adherence status, which allowed for minimisation of bias in the development of core themes. Two other advantages are also worth mentioning were the differential analysis of the adherent and non-adherent group and drawing upon components of social cognitive models. These two approaches have been also been used by other study groups.(424, 425)

Valuable information can be obtained when separating adherent from the non-adherent individuals, potentially identifying strategies for targeting individuals who might be at high risk of non-adherence. Ayow et al.(424) conducted a thematic analysis on eight individual CPAP non-users (<1hr use nightly use over 7 days) and users (>=5hrs in last 7 days). The same themes emerged for both groups: physical (e.g. increased energy, weight loss), psychological (e.g. beliefs about OSA, experienced psychological detriment) and social influences (e.g. support). However the way these concepts influenced CPAP use was different between groups. For example, perceived psychological detriment was reported in users when not using the machine: e.g. guilt when

snoring impacts on the partner. In non-users, psychological detriment was reported when using the machine (e.g. feeling uncomfortable in front of partner with the mask on). The authors concluded that two aspects most strongly impacted on CPAP use, namely peer comparison (in terms of illness severity and daytime functioning) and stigma (feeling stigmatised for not functioning and needing to use a CPAP machine).

One limitation of the study that was not considered by the authors was the unequal time elapsed between initiation and interview between the users (32 weeks after initiation) and non-users (121 weeks after treatment initiation). Factors that emerged in the non-users after 2 ½ years might be inherently different from factors emerging after only 8 months in the users.

The use of a social cognitive model as a theoretical framework for qualitative analysis is useful, as it may facilitate translation into interventions for improving adherence. Tyrrell et al.(425) interviewed patients with OSA who had recently abandoned CPAP. Using the health belief model as a framework, the authors identified that the nine interviewed individuals had a poor understanding of their condition and were unsurprisingly not concerned with their severity and their susceptibility to health consequences of their sleep problem. Additionally, individuals described more barriers to CPAP use than benefits and this was considered one of the catalysts for abandoning the treatment. Considering individuals who have completely stopped using the machine is important, however it is unclear in this study, how these individuals were contacted and identified as 'CPAP abandoners'. A selection bias might have occurred, if individuals, who took part were those more content with admitting that they had stopped using the machine than individuals who refused participation. Arguably these individuals may have displayed a different psychological profile.

In summary, despite the range of study designs, certain commonalities can be identified across these qualitative studies. CPAP is a treatment that is not easily implemented and users find themselves having to utilise internal means such as willpower and self-efficacy often pushing through early resistance to this intervention. Some individuals struggle with the concept of using the CPAP machine, which they believe identifies them as ill and not able to take care of themselves. However, a cost-benefit evaluation whereby advantages (especially the elimination of negative consequences of OSA) outweigh these

disadvantages potentially translates to increased CPAP use, along with social support from spouse/family members and/or health care team. To an extent this overlaps with the quantitative studies outlined in chapter 1 and more importantly supports the transition witnessed in the literature to increasingly focus on psychosocial variables. One limitation of these qualitative studies is generalisability; with small sample sizes conclusions can only be made about particular samples. There is a need to conduct further qualitative studies with similar designs to allow for potential amalgamation at the theoretical level, with the goal of increasing the external validity of some of these study findings.(426)

The aim of this chapter therefore was to investigate qualitatively the experience of CPAP adherence using a longitudinal study design. Similar to Sawyer et al.'s study(423), a short timeframe of 1 week after the CPAP purchase was selected, in addition to a pre-titration and a three months interview. This time line was selected because of the frequent abandonment of the machine within the first month and because adherence patterns are established within the first few days.

3.3 Methods

3.3.1 Participants

Individuals, who had recently been diagnosed with OSA and were committed to a titration study, were contacted for participation. Additional inclusion criteria were: individuals between 18-75 yrs., English language fluency and willingness to comply with the study procedures. Patients who had a co-morbid sleep or psychological disorder were not included. Of 11 individuals recruited, 6 participants were able to commit to three interviews. Two individuals never purchased a machine, one was not contactable after the first interview, and two individuals were only able to attend two of the three interviews. Therefore, all data analysis considered only the homogenous group who were able to complete all three interviews.

3.3.2 Procedure

Ethical approval was obtained by the Sydney Local Health District Ethics Review Committee (RPAH Zone). The consent form and patient information sheet are

presented in Appendices 2 and 3 respectively. Prior to their overnight titration study and upon obtaining consent, eligible OSA patients were invited for the first interview, which was conducted in a familiar environment at the sleep centre. The study procedures were explained and participants were made aware that interviews were audio-recorded with their permission and responses would remain anonymous. Participants were briefed about their ability to withdraw from the study at any time. To make each participant feel comfortable, the interview schedule began with general questions about the history of their sleep problem. Subsequent questions covered the individual's thoughts and feelings about diagnosis and introduction to CPAP, and lastly their expectations of how the treatment might impact on their daily life. Additional prompts standard for semi-structured interviews were included, such as: "Can you tell me more about this", "Can you explain what you mean". Individuals were asked to contact the interviewer after purchasing the CPAP machine. A follow-up phone call was made if the individual had not contacted the interviewer within 2-3 weeks post titration study. The two post titration interviews (1 week and 3 months after purchasing the machine) differed slightly from the baseline interview questions, which were phrased prospectively e.g. "How do you think CPAP will affect your every day life?". The second two interviews included questions about participants' initial experiences with the machine and how they felt about using the mask at the current time. Thereafter the topic of adherence was introduced, individuals were asked to describe a typical night where they used/did not use the machine and what factors they believed influenced adherence. The interview schedule is outlined in Appendix 4. Of note, the interview schedule for this group was the same for the CBT-I group (presented in chapter 4), thus for simplicity reasons, the schedule presented in the appendix is a merged version. The two versions only differed in respect to references to the sleep problem (sleep apnoea/insomnia) or the treatment (CPAP/CBT-I).

The interviewer was not involved in any aspect of the care (diagnosis, titration study or any follow-up sessions); the sleep unit staff, as part of their routine duty, undertook this.

3.3.3 Data Preparation and Analysis

Interviews were transcribed verbatim by the lead researcher, and the data were explored using thematic analysis. This approach to qualitative data has been used extensively, unfortunately often without explicit reference to its use.(427). It has offered a variety of studies- including qualitative explorations of patient adherence(428-430) with a flexible technique that is not restricted by 'pre-existing theoretical framework[s]'.(427, p. 81) An inductive, descriptive approach to analysis was chosen to allow for preliminary exploration of the relatively poorly understood topic of CPAP adherence. According to Braun's methodology outline of thematic analysis,(427) each interview was read and re-read, familiarising oneself with the data set. Subsequently, passages were coded at the basic, descriptive level, and these codes were then collated into themes. The clustering of codes and identification of themes was refined in an iterative process, data extracts were re-read to ensure the themes were internally consistent and captured the verbatim evidence accurately. Lastly, themes were organised into a meaningful account of the individual's experience using sufficient verbatim evidence and telling the story in the words of the participants. A second reviewer warranted the reliability of the data by assessing the analysis at different stages of the process. Themes were also refined by discussions with researchers independent from the analysis process. Participant's names and additional identifying information were changed to ensure anonymity. Small changes were made in the final report to facilitate legibility of the verbal extracts. Brief vocal utterances (e.g. 'ehm' 'oh') were omitted and semantic repetitions (e.g. my partner, you know my partner) or brief tangents from the topic, were also transcribed, but replaced in with '[…]' in the final write up. Short pauses in the interview were transcribed as '…' and incorrect grammar indicated with [sic].

Additional aspects were considered when dealing with the three time points. Participant's three interviews were examined together, to obtain a sense of change within each individual. Emergent themes were then considered across individuals to generate a coherent picture of the experience of CPAP treatment. In order to describe changes across time and allow for a flow in the description of the phenomenon (CPAP adherence), quotes are presented separately, but referred to in the text.

3.4 Results

Patient characteristics of the six individuals who completed all three interviews are presented in Table 7.

Table 7: Patient Characteristics

Alias	Age (yrs)	RDI (events per hr)	Minimum Oxygen Saturation (%)	BMI (kg/m2)	CPAP pressure (cm H_2O)	Marital Status	CPAP use/night
Matthew	38	11	88	33	13	Married	<4hrs
Benedict	40	23.3	89	25.2	10	Married	>6hrs
Stephanie	48	40	81	35	15	Single	4-6hrs
Danny	49	29.6	84	34	9	Live-in partner	missing
Jacob	45	8.5	89	34.5	10	Married	>6hrs
Jim	44	55	71	37	13	Married	4-6hrs
AVERAGE	44	27.9	82.2	33.1	11.6	-	-

From the information provided in the interviews, three themes emerged: Internal conflict around acceptance and implementation, integration of CPAP into life and motivators and resources. Box 2 depicts these three themes and their corresponding subthemes.

Box 2: Overview of themes and subthemes

Theme 1: Internal conflict around acceptance and implementation
Subthemes:
- Being in two minds
- Longing for normality
- Resolving internal conflict

Theme 2: Integration of CPAP into life
Subthemes:
- Integration into nightly routines
- Integration into personal life

Theme 3: Motivators and Resources
Subthemes:
- Intrinsic motivators and resources
- Extrinsic motivators and resources

3.4.1 Theme 1: Internal conflict around acceptance and implementation

In the interviews CPAP was presented as a challenging treatment and individuals were faced with a conflict around acceptance and implementation. Individuals were in two minds, knowing on the one side that the treatment would procure benefits for them, and on the other hand, CPAP was not something they were keen to accept and integrate into their routine. A conflict existed around the concept of longing for normalcy, where CPAP embodied the key to regaining

normal functioning, however was not considered a normal treatment. However individuals developed strategies to facilitate the resolution of these conflicts.

3.4.1.1 Being in two minds

As an artefact of simply being interviewed shortly before or on the day of their CPAP titration study, it was presumed these participants had reached a stage of acceptance. Nevertheless, throughout the interviews, it emerged that this sample was, battling an internal conflict around acceptance and implementation of the machine. Terms used to describe this conflict were having a 'split personality' [Matthew], seeing the one 'side/hand' versus the other [Benedict, Jim], showing 'resistance' despite known advantages of the CPAP machine [Stephanie, Jim]. The latter description is particularly interesting, these individuals were fully aware of the potential benefits of using the machine and risks associated with not doing so, yet just did not want to use the machine (Box 3 quote 1-2, 5). This is particularly pertinent in Stephanie's description of this internal conflict (Box 3, quote 3-4). At three months, a few individuals still described this conflict, highlighting this potential obstacle for adherence persists after these individuals had purchased the machine and were supposedly accepting of treatment. (Box 3, quote 5-6)

Box 3: SUBTHEME-Being in two minds

Being in two minds

1. 'I can see the benefits by having the machine, but I am just, in the back of my mind, will these benefits stay the same, or will my body get used to having this extra oxygen and then become, go back to normal [pre-treatment].' [**Jacob**, 1, l. 200-203]

2. 'Just a change in your life that you don't really want to have to implement. You know that is basically what it is. "I don't have to do this. Why should I have to do this?" You know, but reality is, you have got to do it.' [**Jim**, 2, l. 465-467]

3. 'Its logical, it's rational, and yet I don't want to do it.' [**Stephanie**, 1, l. 273-274]

4. 'I have to do something about my sleep, so, even if I am negative about it, I still have to control the feelings.' [**Stephanie**, 1, l. 167-168]

5. 'How it makes me feel is fantastic, but how it makes me feel when I'm wearing it is not so great, and it's that sort of thing…all the way along.' [**Danny**, 3, l. 247-249]

6. 'I try to avoid routine where I can because it's boring, you know but that is on the one hand, but on the other hand this routine is not that much of a problem for me, this routine shows me benefits you know. So I am almost a split personality when it comes to this.' [**Jim**, 3, l. 435-438]

3.4.1.2 Longing for normality

The debilitating daytime consequences of sleep apnoea had proven an obstruction to normality in the majority of individuals.

'what a normal person…what used to take me an hour to do in the mornings, it would probably take me a couple of hours to do now.' [Jim, 1, l.39-41]

'I don't like the sensation of just you know, one minute being you know… reading the newspaper or something and then "boom" you are gone, you know sort of, fall fast asleep.' [Stephanie, 1, l. 189-191]

'there are some days where you get up and you really just cant get going, […] and as a result you waste a day, […] you end up in front of the telly and doing all the things that you shouldn't be doing instead of getting on with life.' [Danny, 1, l.56-60]

CPAP presented itself as the key to regaining a sense of normality (Box 4, quotes 1-6). Although the treatment was considered as something that would aid with the two most basic processes: sleep and breathing. Being unable to do this independently, portrayed the individual as abnormal. (Box 4, quotes 7-15)

It was evident across time points that individuals longed for this sense of normality. At the first interview individuals were hopeful in reaching this goal by using the CPAP machine (Box 4, quote 1-2), whilst remaining realistic, like Danny, who judged normality by what he was able to do prior to developing OSA symptoms (Box 4, quote 3). During the second interview, and even more so after 3 months, individuals began to identify a gradual progression to regaining normality during the day (Box 4, quotes 4-6). In conflict with this development was the sense of wearing the machine at night was not considered normal; CPAP was identified as a treatment for a sleep related breathing disorder. Sleep and breathing present two of the most basic human processes. Acceptance and use of the machine was equated with being unable to sleep and breathe independently, something that set them apart from 'normal' people (Box 4, quotes 7-10). There was a certain stigma associated with this, and individuals identified themselves as 'old and sick', akin to an 'invalid'[Jim], and as if the body was 'giving up' [Danny]. These terms reflected their own representations; individuals also feared the stigmatisation by others (Box 4, quotes 11-12), particularly Stephanie voiced her trepidation of accepting the machine, assuming others, especially work colleagues, would judge her as not be able to function normally.

Box 4: SUBTHEME- Longing for normality

Longing for normality

1. 'Yeah, hopefully like it will work for me and yeah, like it becomes all good again.' [**Matthew**, 1, l. 241]

2. 'If I get, the benefit, the full benefit that I envisage from this thing, I am going to have part of my life back, that I haven't had that...haven't had for some time.' [**Jim** 1, l. 234-236]

3. 'I am not expecting to maybe [...], run a marathon or something, but I want to have some resemblance of normality back in my life.' [**Danny**, 1, l. 328-330]

4. 'this is a bit like a time warp [...] Back to when it was...to what it was before, or how I felt before, I thought that was the norm, but then no it's not.' [**Danny**, 3, l. 113-118]

5. 'Like just happy and feeling...get back, like just happy, happy that I got it and thinking of getting back on track.' [**Matthew**, 3, l. 236-237]

6. 'yeah I have got sleep apnoea, I have still...but by me having the machine, I am back to normal.' [**Jacob**, 2, l. 443-44]

7. 'I have something determining my sleeping, you know, how I sleep. You know there is ...and that is, I have got to follow a particular routine to get to go to sleep [emphasis].' [**Jim**, 1, l.186-188]

8. 'Its like you loose your independence as a human being, like you need this machine to breathe, you are artificially living.' [**Jacob**, 1, l. 281-282]

9. 'it's just a stigma, you know, you don't want to feel that your body is giving up on you, or you need assistance, from anybody, especially if you are doing something as simple as breathing, we are all supposed to be able to do that.' [**Danny**, 1, l. 262-264]

10. 'you are doing something that, your doing something that the rest of the population is not doing. So, yes, sort of disappointing from that aspect.' [**Benedict**, 3, l. 59-60];

11. 'like "there is something wrong with him, he has got a machine to breathe you know".' [**Jacob**, 1, l. 240]

12. 'I am a professional person doing a professional job, and I don't want to be judged as somehow not able to do my job properly.' [**Stephanie**, 1, l. 347-349]

13. 'It is not nice, [...] at the end of the day, you know, it is a machine, it is an artificial thing. It is still in the back of your head, you know, then you are getting the great benefits, you can [emphasis] sleep, you can [emphasis] tolerate the noise, you can [emphasis] tolerate the heat from the strap; you are feeling good, energy...but why can't you do it by yourself, naturally.' [**Jacob**, 2, l.715-719]

14. 'It [CPAP] doesn't make me feel any different about who I am, other than in that one...that one aspect [...] I don't know... father figure, man figure...is that you can gather her, flop to your chest and hold them and make everything better, and I just feel like this gets...in a way a little bit of that.' [**Danny** 3, l. 380-384]

15. 'At one stage I was thinking 'if I use this machine for too long, is it going to weaken my ability to breathe on my own?' [**Jacob**, 3, l.224-225]

Interestingly, these concerns were still present at the 1-week and 3 month interviews, suggesting these concerns might be contributing somewhat to the levels of CPAP adherence in these individuals (Box 4, quotes 13-15).

Seemingly, there is uniqueness about CPAP as a treatment for a sleep related breathing disorder. Potentially more so than other disorders, the sense of abnormality of receiving treatment is increased by CPAP being a treatment for two most basic physiological processes. A quote by Danny highlights how this contributes to his internal conflict:

'I don't like to feel that I am, you know like I can't look after myself. And you know even with the diabetes you look after yourself because you keep your blood sugar levels, so you monitor your blood sugar levels, […] but this you are not, essentially it's a bit different, you are not really in control of this.' [Danny, 1, l. 269-275]

3.4.1.3 Resolving internal conflict

Individuals described different strategies for overcoming this internal conflict. For Jim, the process of simply speaking openly about it during the interviews was sufficient to crystallise the root of his resistance and resolve it (Box 5, quote 1), for others, an effective method was acceptance, being rational about the need for change or simply placing mind over matter (Box 5, quotes 2-4). At the second and third interview these strategies became more concrete. Considering how the benefits of using the machine outweighed the disadvantages for change was reported by a number of individuals (Box 5, quotes 5-7). Drawing social comparisons was also important, particularly for Jim (Box 5, quote 8), who compared his conflict with others who have been through the same process. Interestingly, when evaluating the disadvantages against the benefits (Box 5, quote 5), it became clear how Jim resolved the conflict around feeling like an 'invalid' (see above): understanding that treatment did not necessarily equate to a poor quality of life was a substantial discovery for him.

There is an implicit assumption in this subtheme, that the resolution of this conflict is based on a rational decision-making process. However, there are a number of somewhat unconscious processes revealed in the interviews. Firstly, a concept borrowed from social psychology: cognitive dissonance; a concept that describes the simultaneous presence of two conflicting beliefs; individuals have a tendency to want to resolve this dissonance by changing the balance between

these two conflicting beliefs. The resolution of conflict, according to the theory, occurs largely in the unconscious.(431) This was the case for both Matthew and Stephanie (Box 5, quotes 9-10), who seemingly reduced dissonance by attributing greater importance to the potential health benefits (compared to the disadvantages for change). For Matthew this was accomplished by being conscious of the money invested into purchasing the machine; for Stephanie it was the belief in her level of intelligence. Secondly, it is important to note that the adherence behaviour of interest here occurs during sleep, when certain stages are associated with reduced activity in rational/decision making regions of the brain, and activity in emotional regions is increased. Waking from REM sleep might result non-adherence behaviour (i.e., taking off the mask), which is precisely not based on rational decision-making processes. This phenomenon was reported by two of the six participants in this sample (Box 5, quotes 11-12).

Box 5: SUBTHEME- Resolving conflict

Resolving Conflict

1. 'I mean talking about therapy on an emotional level, well it served a great purpose on that regard [breaking through resistance]. I guess I wouldn't really have crystallised it, had I not had this interview with you.' [**Jim**, 2, l. 565-568]

2. 'But life isn't fair anyway as life is, some people are more, their health is better than the other people's health you know what I mean? But, it's at the end of the day, everybody gets fixed up, it's the main thing you know. That is how life is you know.' [**Matthew**, 1, l. 414-417],

3. 'I need the treatment to have better sleep so that I can function better, so. I'll just get on with it. I mean, that just...you have got to rationally face it.' [**Stephanie**, 1, l. 678-680]

4. 'So it is definitely there, the downside that is part of the downside. But, I can't hang around thinking; you know thinking about that for too long.' [**Jim**, 2, l. 198-200]

5. 'For me just to feel OK about doing this, it's a relief to have broken through that resistance, It's a great reminder to take care of my health. It's made me realise there are probably a lot of people out there [...] these people are just carrying on with their lives. It makes me realise that the person in a wheelchair who is full of life, is living a more fulfilled life that a lot of other people.' [**Jim**, 2, l.552-559]

6. 'It's more detrimental if I don't use it, than if I use it. So the benefits and the safety are more important than me feeling, you know, a little bit uncomfortable in wearing it initially.' [**Jacob**, 3, l. 336-339]

7. 'The feeling of CPAP being unnatural], it's OK for me, so long as I can see that the, the health benefits that I thought of is going to get.' [**Benedict**, 3, l. 243-244]

8. 'There are people that do have problems with it, and it was interesting for me to hear that, considering my experience to date with it all. I was kind of "I am not alone you know if taking so long to get here. I am not alone in resisting it", made me feel a bit better about myself.' [**Jim**, 2, l. 482-486]

9. 'To use the machine? If I didn't want the help I wouldn't come here. If I didn't want the help, from the doctor I wouldn't come here and spend my money.' [**Matthew**, 2, l. 356-357]

10. 'If you know you have a condition and you have therapy that can alleviate it and potentially make sure you live an OK length of life, rather than a shorter length of life, you're a bit stupid if you don't do it. But I don't think I'm stupid, so I persist.' [**Stephanie**, 3, l. 537-540]

11. 'There's not a rational thinking process going on there. It's...ahm, you know its an un... it's uncomfortable and that's the reaction I'm going for [...] I don't think I'm really awake enough to [think rationally].' [**Danny**, 2, l. 684-690]

12. 'Oh, it happens every night! I take it off every night. For me not to take it off is quite rare. [...] Sometimes I remember taking it off, but most times I don't.' [**Jim**, 3, l. 269-281]

3.4.2 Theme 2: Integration of CPAP into life

At the first interview, individuals had little conceptualisation of the impact CPAP would have on their lives. Many raised questions and described expectations

they had. For some, their predictions materialised, whereas for others they did not. The titration study offered the primary opportunity to experience the CPAP machine and what it would mean to integrate this treatment into their life. Some described this experience as *'the worst night sleep'* [Jim] or feeling *'jetlagged'* [Stephanie], with no beneficial effect of using the machine upon awakening, whereas others again witnessed immediate benefits:

'I felt that I didn't sleep for a long time, but I have [sic] a euphoric experience the next day basically, I felt like on top of the world. I felt like, wow. "Wow pow catch me if you can." I was like, "I am heaven" basically, "cloud nine," I felt really good.' [Jacob, 2, l. 104-106]

Apart from Jim, none of the individuals expressed a positive or negative impact of the titration study experience on subsequent machine purchase:

'it took me a month to get to that point of going and getting the machine and I was more ready a month later than I was the day after getting the prescription, or the day I received the prescription, so I felt ready to try it out.' [Jim, 2, l. 63-66]

Benedict stated a more detailed focus on the practical experience of CPAP on the titration study night would have helped him form most positive or helpful expectations around integrating CPAP into his life:

'a bit more time in the education session with the practical stuff about the mask fitting and getting used to the machine. I think that's, the more time that you have with the machine before you actually go home is a good thing, so you know what to expect.' [Benedict, 2, l. 236-238]

Throughout the interviews, individual's described their initial expectations and subsequent experience of the integration of CPAP into their life, both at a night-to-night, but also at a more general and personal level, as well as considering the impact on family/relationships and their future:

3.4.2.1 Integration into nightly routine

Individuals expressed the need to integrate CPAP into their everyday nightly routine, moving towards a stage of *'familiarity'* [Benedict], with the hope that the therapy would eventually become *'second nature'* [Stephanie], *almost 'like wearing glasses'* [Jim] or having *'a ring on your finger'* [Matthew]. They described the need to persist until the new behaviour became a habit (Box 6; quote 1, 2). Strategies to facilitate this process such as gradual nightly increase

in use or anchoring it to existing routines were employed by Matthew and Danny respectively (Box 6, quote 3, 4). One particular concern during this process however was the potential impact on sleep and the bedtime routine. Being accustomed to falling asleep within a few minutes, individuals were apprehensive at the baseline interview about the impact this treatment would have on their sleep onset (Box 6, quote 5). Interestingly some individuals (e.g. Box 6- quote 6, 7) noted the increase in sleep onset latency after using the machine at the 1-and 3-month interview. In addition to changes in actual sleep, the bedtime routine was affected by the integration of the CPAP machine into daily life. Not being able to read a book, have a conversation or be intimate with the bed partner were adaptations these frustrated individuals had to make. This was not confined to their own routine, as described by Jacob (Box 6; quote 8), but concerns extended to the impact on the routine shared with the bed partner (Box 6; quote 9).

Box 6: SUBTHEME - Integration of CPAP into nightly routines

Integration of CPAP into nightly routines

1. 'I suppose it's just hoping it just becomes second nature. Part of your routine at [sic] going to bed at night is putting that thing on.' [**Stephanie**, 1, l. 451-453]

2. 'getting used to it was uncomfortable at first, so I knew that was going to, you know I'd have to adjust to the sensation and the feeling of having a mask both in the face and the air going up your nose.' [**Benedict**, 3, l. 147-149]

3. 'Yeah, I would put it on the first couple of hours, that is how much I could do. I couldn't do anymore than that and that is why I took it off. The second day, I did it 4-5 hours, that is as much as I could handle. Its good though the 4-5 hours, and then I have a break and then I put it on again for the next couple of hours.' [**Matthew**, 2, l. 399-402]

4. 'From a daily ahm, point of view, I've just tried to sort of incorporate it. When we bath the baby, that's when I sort of bring the stuff out, the machine, I use that as the reminder to take the things out.' [**Danny**, 3, l. 343-346]

5. 'I don't know how uncomfortable its going to be, or whether its going to hinder me actually getting to sleep.' [**Bendict**, 1, l. 166-168]

6. 'Like I like to fall asleep in 5 minutes, bang, like I...but I am slowly getting to that its taking me like 15-20 minutes to fall asleep.' [**Jacob**, 2, l. 141-142]

7. 'I have really noticed in the 3 months, it takes me a lot longer to go to sleep than it did in the past. But as people pointed out, in the past I was actually buggered, so not surprising I dropped off in 2 seconds.' [**Stephanie**, 3, l. 251-254]

8. 'you want to go to the toilet, it's a pain in the backside, you understand. You want to get up and say read something, you have got to unhook yourself [laughs], so its, you are in a way locked in sort of situation.' [**Jacob**, 2, l. 88-90]

9. 'Usually I go to bed after her [partner], when she has fallen asleep, so now if I am going to put this thing on and hook this on, and switch it on, I don't know what I am going to do. Might have to do with an extra bedroom.' [**Matthew**, 1, l. 177-180]

10. 'it's actually seems to have settled down a bit now, but the skin here [points to part on nose] has been quite irritating. Ahm, and I've actually...I've just put it in the draw and I've done without it for the last couple of hours.' [**Danny** 2, l. 470-472]

11. 'sometimes you have got air blowing in your eye, which isn't particularly comfortable, you know, so you have got to adjust the mask and it is just a hassle,[...] and you are back to having broken sleep you know.' [**Jim**, 2, l. 336-339]

The integration of CPAP was accompanied by unwanted side effects in some individuals. Most commonly reported effects were skin irritations, air leakages and marks from the straps (Box 6; quotes 10,11). This experience may

have impacted on CPAP use in the short term, with some individuals reporting not using the machine on a few nights as a result. However, generally, these individuals re-initiated use by adopting active coping styles such as speaking to the CPAP distributor or nurse, or self-experimenting with the mask and machine settings.

3.4.2.2 Integration into personal life

In addition to incorporating treatment into their nightly routines, individuals described the process of integration at a more general and personal level. How does CPAP impact on the family and personal relationships? How does CPAP fit into the personal outlook? What personal circumstances make integration difficult? Before the titration night, concerns were voiced about the potential impact of CPAP on family life and new or existing romantic relationships (Box 7; quotes 1,2). These were either practical implications, such as the noise of the machine disrupting the partner's sleep or more intimate issues, such as fear of embarrassment or interference with closeness. For some, like Jim (Box 7, quote 3) the concerns did not materialise and CPAP was seamlessly integrated into his personal life. Yet for others, even at 3 months the introduction of CPAP resulted in sleeping in separate rooms to their partner (Box 7, quote 4), or the machine was something that prohibited intimacy.

Individuals also evaluated CPAP in relation to the future. For the majority, at the baseline interview and even at week 1, CPAP embodied a treatment that would be integrated in the short term; it was described as a crutch, a technique that would help them regain the necessary energy and motivation to lose weight and no longer require the machine (Box 7, quotes 5-7). Gradually over the 3 months, individuals began to accept that CPAP was a treatment for life (Box 7, quotes 8-9) and would require long-term integration. For some this step towards acceptance was hindered by their age and the thought of implementing this treatment for the next 25-30 years (Box 7, quote 10).

Box 7: SUBTHEME- Integration of CPAP into personal life

Integration of CPAP into personal life

1. 'We don't have a bogeyman, but maybe there is a bogeyman about to move in you know. Kids get freaked out by different things and I am concerned that that may scare them a little bit.' [**Jim**, 1, l. 199-201]

2. 'Luckily I am single, I don't know how it goes, I suppose, the other partners must be understanding about the partner needing it, I would hate to be young and getting it, because that would really be a passion killer I'd say.' [**Stephanie**, 1, l. 437-440]

3. 'The fact that I can sleep in the same bed as my wife, and my kids are OK, if anything they tease me about it, [...] And it wasn't possibly wasn't as bad as I thought it was going to be.' [**Jim**, 3, 85-91]

4. 'We sleep in separate rooms, [...] it can be a problem, if I sleep in the same bed with her, because the way I have got it hooked up and I, I wanna [sic] sleep on my side,[...] not all the time, but I turn over, and this thing is like rattling on the bedhead.' [**Jacob**, 3, l. 360-365]

5. 'In the back of my positive mind, this is not a life-time commitment for me, even though I have been told by the doctor that it is.' [**Jim**, 1, l. 114-115]

6. 'I am still not 100% sure how long you have to be attached to this thing, months, years, days weeks, or I don't know.' [**Danny**, 1, l. 249-250]

7. 'I'm assuming that I will get to a stage where I will loose some weight and I will not have to use it, ahm every day.' [**Danny**, 2, l. 422-423]

8. 'I am getting this message that this is probably a life thing, you know.' [**Stephanie**, 2, l. 675-676]

9. 'And that is possibly a case of optimism on my part, that I won't have to use the machine anymore [...] maybe it was just a bit of a fantasy.' [**Jim**, 3, 71-73]

10. 'they should somehow motivate people, not only to use it, how to get off it, [...] especially at 45, you don't want to be sleeping on a machine for the next 25 years. [**Jacob**, 2, l. 711-715]

11. 'I think one night when I didn't use it because I had a heavy cold and I had such a shocking night sleep.' [**Stephanie**, 3, l. 235-236]

12. 'We were out late at a party, I think I got home about 2 or 3 in the morning, [...]I thought [...] "it's not worth it".' [**Benedict**, 3, l. 412-414]

Unusual personal circumstances provided additional obstacles for this process of integration. Travel was frequently noted as a concern in the baseline interviews and those who did travel during the first 3 months, noted difficulties using the machine. Two other frequently reported events were the presence of flu symptoms and shorter nights as a result of social occasions (Box 7, quotes 11-

12). Despite these concerns and difficulties experienced, for the majority of the sample this process of integration did not seem too onerous, especially at the three-month interview. Certainly by this point, individuals described having become habituated to using the CPAP machine and integrated it well into their life:

'I don't see it as a hardship; if that's what you're trying to sort of get at. It's the necessary evil.' [Danny, 2, l. 285-286]

'it's not inconveniencing me in doing what I want to do.' [Benedict, 3, l. 351-352]

'That's me, you know, that's who I am, I come with the machine and mask' [Stephanie, 3, l. 819-820]

3.4.3 Theme 3: Motivators and Resources

Individuals described how certain aspects of their sleep problem and potential of CPAP in producing benefits for them (internal) and their spouse/family (external) embodied motivators for adherence. These goals could be obtained by exploiting certain internal and external resources.

3.4.3.1 Intrinsic Motivators and Resources

Individuals at all these interviews reflected on the impact their sleep problem had on their sleep and daytime functioning and how this motivated them to seek treatment. For example sleep apnoea resulted in poor sleep quality, increased sleepiness or reduced social functioning (Box 8, quotes 1-3). Improvements in daytime symptoms such as sleepiness or reduced energy/motivation were significant motivators for continued CPAP use (Box 8, quotes 4-5). Additional changes were also witnessed in the following domains: cognition, personality, social functioning, road safety, performance, nocturnal symptoms, mood and social activities (Box 8, quote 6-7). Apart from these specific changes, individuals also reflected on potential improvements in the foreseeable future, such as the possibility of enjoying a fulfilled life of good health and wellbeing (Box 8, quote 8), in addition to being able to share experiences with family, friends and even new arrivals as in Danny's case (Box 8, quote 9). Moving back into the same bed with the partner after a long history of separate rooms, was one of these shared experiences individuals longed for. These improvements were valued in light of the severity of the sleep problem; some participants

described being motivated by how severe they (and others) rated their illness (Box 8, quote 10).

Participants reported accessing a variety of internal resources in order to motivate themselves to use the CPAP machine and to gain the above-mentioned benefits. Adopting an active coping style was described by a number of individuals. Rather than letting frustration with the machine become too overpowering, leading to non-adherence or even abandonment of the machine; individuals reported using certain strategies to facilitate implementation of CPAP. For example experimenting with the machine settings, contacting the CPAP nurse for support and delaying the bedtime to increase sleepiness were processes that facilitated acclimatisation to the machine (Box 8, quotes 11-13).

Box 8: SUBTHEME - Intrinsic motivators and resources

Intrinsic resources and motivators

1. 'Because of my snoring my quality of sleep is not that great, so I had come along and have a sleep study, you know that is what prompted it.' [**Jim**, 1, l. 14-16]

2. 'It's a problem, because I feel tired, and I shouldn't be that tired, you know basically, that is the bottom line.' [**Jacob**, 1, 457-458]

3. 'I paid the consequences outside of my working day, so at night and weekends I think, the build-up over a week, that's when it whammied at me, and so I think probably more had an impact socially rather than work wise.' [**Stephanie**, 3, l. 102-105]

4. 'I have been using it for a week. And I haven't got this siesta feeling anymore.' [**Jacob**, 2, l. 130-131]

5. 'There's a few incentives there, but the biggest one is that it works, it's not a waste of time, it's not just doing something for the sake of doing something. It genuinely seems to do what it's supposed to do, and that's...give you back you know, some zip, some energy.' [**Danny**, 2, l. 233-237]

6. 'If the results are good, I will feel more energetic and all that sort of stuff, its just I think its going to affect my relationship maybe with my wife a little bit, or I feel it might. I don't know, I am not a 100% about that yet. But if I feel more alert, not as tired, I think it will be good. Good for work, good for motivation to do exercise, to do things. Because at the moment, [...] I can't do anything.' [**Jacob**, 1, l. 464-470]

7. 'Before I start [sic] doing the therapy, I could wake anytime from...see that is another symptom that's alleviated. And I think somebody here was telling me it's probably because I was always in that shallow sleep, anything going on in my body would disrupt me, and I found I was getting up and going to the loo, like anywhere from 1-2 times to 4 times a night. That's totally gone.' [**Stephanie**, 3, l. 627-631]

8. 'Its not as much the snoring for me, because that doesn't effect me, it affects others. But the health benefits, particularly the blood pressure, are the things were, its all driving me to keep going with it.' [**Benedict**, 1, l. 235-237]

9. 'I want to see my son grow up, if I want to enjoy our...you know the relationship for you know many years to come, that's what I need to do.' [**Danny**, 3, l. 168-170]

10. 'If I was somebody who had only had mild things, I think I'd probably be more questioning. Whereas I know from what people have said "you have got to do something about it- its serious," I will do something about it.' [**Stephanie**, 2, l. 736-739]

11. 'I was telling him [CPAP using friend] about what I had discovered about different machines I go "why don't you go try a different machine" da, da, da. So basically he hasn't done that. So he hasn't gone to experiment, to try to find a solution to the problem.' [**Jacob**, 2, l. 601-604]

12. 'The first one [CPAP machine] I probably wouldn't use it, not because I didn't want to use it, I just couldn't do it, and then I went to the clinic, where there were renting them out, I told the person there, to give me like a better one.' [**Matthew**, 2, l. 385-387]

13. 'Normally I go to bed 10-10.30 and I think I have been staying up later to try and make myself be tired by the time I go to bed. So that, to try and help them more quickly falling asleep.' [**Stephanie**, 2, l. 233-235]

14. 'I think at the end of the day, they can help you, but it is up to you probably to help yourself.' [**Matthew**, 3, l. 735-737]

15. 'like I got frustrated the first few nights, whatever, but I am still giving it my best shot, still want to do it.' [**Matthew**, 2, l. 335-336]

16. 'that's what I took it from the medical practitioners the need to keep, you know get help if you need it, but keep at it.' [**Benedict**, 2, l. 274-275]

In these quotes, it becomes apparent that individuals desired active engagement in the treatment, rather than blindly following the instructions of the physician (Box 8, quote 14). However, some individuals described an element of passivity by implementing what they had 'been told' to do [Jim].

Lastly, participants reported intrinsic traits such as willpower and perseverance that could provide the necessary resource for continuing to use the CPAP machine. (Box 8, quotes 15-16)

3.4.3.2 Extrinsic Motivators and Resources

CPAP is a treatment that does not solely involve the individual patient, but is an intervention that needs to be integrated within the family context (see the above theme 'integration of CPAP into personal life'), thus extrinsic motivators and resources are naturally assumed to influence adherent behaviour. This was evident in the present sample. The impact of OSA on the family and close relationships was a large motivator for these individuals to seek and adhere to treatment. Participants wanted rid of both night time (e.g. snoring) and daytime (e.g. sleepiness, irritability) symptoms that were affecting the individual's partners, family or friends (Box 9, quotes 1-2) and in Jacob's case the public (Box 9, quote 3). Witnessed and potential further improvements in these domains were considered and played a role in motivating these individuals (Box 9, quotes 4-6).

Box 9: SUBTHEME - Extrinsic Motivators and Resources

Extrinsic motivators and Resources

1. 'We went out one time and I fell asleep in the restaurant. I was tired, I mean I had been up all night and all day and out in the evening and... but that shouldn't be happening, its really embarrassing her, and I don't want to embarrass her, I want that to be gone.' [**Danny**, 1, l. 325-328]

2. 'That is something I am always conscious of when I go away with friends, is that I know I snore loudly. And all that sort of thing, so....sometimes that does put me off.' [**Stephanie**, 1, l. 107-109]

3. 'So that I don't feel tired and don't become a danger to the public and myself, when driving.' [**Jacob**, 3, l. 436-437]

4. 'just be able to I suppose just lie and have a chat with my wife in bed is a nice thing rather than her having to go to sleep before me, you know because of my snoring.' [**Jim**, 2, l. 211-213]

5. 'She will probably be happier if I am not as grumpy, you know if I am not as tired. If those things improve I think she will be happy in that respect.' [**Jacob**, 1, l. 189-190]

6. 'that's also having benefits for those other people, so you know my wife doesn't have to hear me ah snore as loud. My wife doesn't have to hear me gasping for breath. So I feel good about that.' [**Benedict**, 3, l. 335-338]

7. 'It feels good that she is standing by me you know. She is the one that wanted me to do all this kind of stuff. Yeah, cos [sic] I would probably, I wouldn't...not think about doing it you know.' [**Matthew**, 2, l. 77-179]

8. 'My sister has been brilliant, she has been ringing me on a regular basis saying "how're we doing" and she talks about what she does, or she gives me suggestions like with the air blowing on my face, she said "oh well try turning the tube up over your head".' [**Stephanie**, 2, 319-323]

9. 'The staff here have been great, you know taking the time that has been spent here, you know, I was comfortable doing it, so that was great. And they helped you pretty well every step of the way.' [**Jim**, 2, l. 641-643]

10. 'I think they should be doing more, or better fitting of the masks, or really, really walking you through with the mask. Because you don't really...when you first start using it, you don't really know if you've fitting it right.' [**Stephanie**, 2, l. 215-218]

11. 'I think it would have been better to spend less time in that session in before hand about what sleep apnoea is, because I think we covered that, particularly in the first two consultations with the clinicians and more time on saying, you know this is what it involves.[....] And maybe, you know some tips about how to help you, you know stick at it, so to speak, particularly in the old days.' [**Benedict**, 3, l. 111-122]

12. 'I think it's sort of been like "this is your diagnosis, this is what the treatment is" its not like [...] "how can we keep you on this, or what can we do to help you be on this treatment".' [**Stephanie**, 2, l. 856-859]

13. 'I feel in that respect I feel I have had better follow-up or more care, if I can use that word, for the person I bought the machine from, then from here (sleep centre).' [**Benedict**, 2, l. 289-291]

14. 'I got married about 5 years ago, ahm, it was getting worse and worse, and the first 3-4 years, she could cope with it, about 2 year she couldn't cope no more. "Yeah, fix yourself up, or I leave you" [laughs].' [**Matthew**, 1, l. 42-44]

15. 'It is not always good to push me, because then I will just say "oh stuff that, I don't want to do it". So I...probably I did resent being pushed.' [**Stephanie**, 1, l. 67-68]

16. 'He [physician] basically said this is it and that is it. Basically he was pretty blunt about it, I am a bit different, [...] I explain things to them a bit more compassionately.' [**Jacob**, 1, l. 326-332].

Extrinsic resources were available to these individuals. Support obtained from the spouse/family was reported as advantageous for increased CPAP use

(Box 9, quote 7). Support from the medical team (physician, CPAP nurse, contact at CPAP distributor) or peers (other CPAP users) were vital for troubleshooting as well as emotional support (Box 9, quotes 8-9), however some rated the input from the medical team below their expectations. Stephanie and Benedict for example desired increased opportunity during the education session (prior to the titration study) for getting to grips with the practicalities of the machine (Box 9, quotes 10-11). There was a consensus amongst these two individuals that there was increased focus on sleep education and less on concrete implementation of the treatment (Box 9, quotes 11-12). For Benedict this type of support was optimally provided from the CPAP distributor (Box 9, quotes 13). It is important to note that for some individuals there was a fine line between support and external pressure. Well-meant support from the spouse could often be interpreted as pressure, described by Matthew in a joking manner (Box 9, quote 14), however for Stephanie the pressure experienced from her mother was more serious situation and actually led to increased resistance towards seeking treatment (Box 9, quote 15). Jacob underwent a similar experience from the medical team (Box 9, quote 16) envisaging a more co-operative, patient-centred style than was offered by the physician.

3.5 Discussion

CPAP, the current treatment of choice for OSA is undoubtedly a demanding intrusive treatment. Although the literature has predominantly focused on biomedical factors as discussed in chapter one and two, a handful of studies has investigated psychosocial variables using both quantitative and qualitative methodologies. The qualitative studies have drawn upon a wide variety of methodologies and analyses and themes emerging in these studies can be compared to the results reported here. This chapter provides a longitudinal perspective upon CPAP use, highlighting important experiences from pre-titration through 1 week and 3 months post-treatment initiation. Figure 6 is a visual representation summarising the emerging themes; how they relate to one another is explained below.

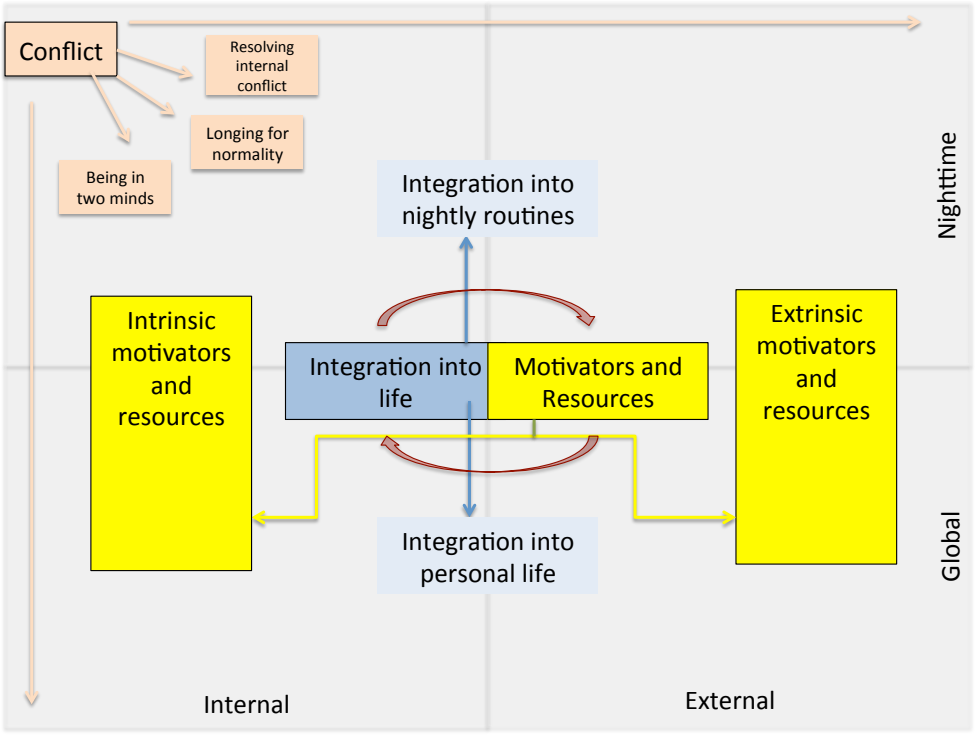


Figure 6: Summary of Results

3.5.1 Conflict

The results presented in this chapter highlight how individuals experience a conflict around initial acceptance and subsequent adherence to the machine, describing a state of being in two minds, influenced by their knowledge of OSA and its consequences, and on the other hand being held back by simply not wanting to use the machine. What individuals longed for was to obtain a sense of normality; which often created further conflict as CPAP is not a normal

treatment, but is the key to normal functioning. This conflict was less evident in the 3-month interview, potentially as a result of the conflict being resolved, using various different strategies, such as focusing on the benefits or acceptance of the situation. This theme highlights how CPAP adherence can be a very dynamic process, with individuals potentially moving back and forth between seeing the necessity for change, but also being reluctant to engage in change. Some individuals seem to resolve this conflict early on and succeed in integrating the treatment into their lives; others are delayed and even at 3 months after purchasing the machine still describe a certain resistance to using the machine. A model that has acknowledged this very dynamic process is the transtheoretical model. This theory (described further in chapter 6) states individuals pass through five distinct stages of change, and throughout this transition weigh up the pros and cons for change. Their belief in their abilities to make the behaviour change (self-efficacy) can vary as a function of the transition through these stages, with self-efficacy being increased in the theoretically higher stages. Lastly, the transition is facilitated by the adoption of certain strategies; cognitive approaches encourage movement through the earliest stages whilst behavioural approaches predominate in the transition through the later stages.

This assumption differs significantly from the view commonly adopted in the literature that change (i.e. adherence) is a static entity. Earlier reports dichotomised CPAP adherence using the cut-off of 4hrs on 70% of nights(181) and evidence for the stability of these dichotomous patterns after only a few days.(203, 204) However, a recent time-series analysis of 71 CPAP users over 365 days, revealed seven behavioural patterns, five of which were identified as non-stable (slow improvers, slow decliners, variable users, occasional attempters and early dropouts), supporting the notion that adherence is a dynamic process.(205) Additionally, the components of the transtheoretical model assessed early in the treatment stages predict subsequent CPAP use(237, 286, 287) and motivational interviewing/enhancement therapy (MI/ME) has indicated improvements in CPAP use, with one study reporting small to moderate effect sizes(Cohen's d of 0.3-0.6(354) and another study revealing a 2hr increase in median nightly CPAP use in the intervention arm compared to the placebo control group at 12 months.(355) MI/ME approaches acknowledge that some individuals will present with ambivalence towards change and require the therapist to 'role with resistance'.(432, 433)

In this theme, it also emerged that CPAP embodied an obstruction of normality of two basic processes: sleeping and breathing. Individuals described feeling stigmatised by others and to an extent internalising these beliefs themselves. CPAP as a treatment for a sleep related breathing disorder was equivalent to showing vulnerability, feeling old or sick. Both an internal(419) and external(424) stigmatisation has been reported by other qualitative studies. This highlights the increased importance of psychosocial interventions in improving CPAP adherence. Wearing a machine every night to go to sleep is different from simply 'popping a pill', which is indisputable. However, it might also differ from other complex treatment interventions. As explained by one of the participants, CPAP was different from controlling his blood sugar levels for his diabetes. The question arises, is there something unique about this treatment for a sleeping and breathing related problem that requires specific attention over and above what is addressed through basic medical interventions. This aspect has not been explicitly targeted by any of the existing interventions. One study interested in the effects of cognitive behavioural therapy in increasing CPAP use, did however include role models (existing CPAP users), who described their positive (and negative) experience with the CPAP machine, emphasised the potential benefits of adhering to the treatment and the importance of pushing through early resistance.(351) The intervention devised by Richards and colleagues took part in a group format including up to 10 individuals and their partners. These aspects although not explicitly targeting stigma, might have effectively reduced the individual's and partner's dysfunctional beliefs about the CPAP machine. Future interventions aimed at increasing CPAP adherence might benefit from a) explicit targeting of potential stigma of using the machine b) inclusion of spouse/important family member c) include other CPAP users in a group format to somewhat normalise the CPAP experience.(see 416)

The participants in this study also described using techniques to resolve the conflict they faced. One strategy that was mentioned by a number of participants was to focus on the benefits CPAP provided. This sense of weighing up the advantages against the disadvantages of change emerged in numerous qualitative studies described in the introduction.(421, 423-425) This cost-benefit evaluation has also been described in some quantitative studies that have evaluated this process as a component of social cognitive and behaviour change models (e.g. health belief model, transtheoretical model). The health belief

model for example describes how individuals engage in a cost benefit evaluation '*wherein the individual weighs the action's effectiveness against perceptions that it may be expensive, dangerous (e.g., side effects, iatrogenic outcomes), unpleasant (e.g., painful, difficult, upsetting), inconvenient, time-consuming, and so forth'*(434, p. 2) and the transtheoretical model explains how a balance between the pros and cons differ at different stages of the change process (decisional balance). Additionally, the process of consciousness raising, a cognitive strategy that facilitates the transition through the earliest stages of change, may help individuals focus on the benefits and how these outweigh the costs.

A second strategy to resolve this conflict involved acceptance of the need for treatment, despite its difficulties. Acceptance and commitment therapy (ACT) can be considered a third-wave therapy successful in producing significant improvements for a variety of psychological disorders.(435) Although not extensively adopted for the use of improving patient adherence, and not at all investigated as a potential intervention for CPAP use, some attempts have been made to investigate the usefulness of ACT in improving patient adherence. For example, a recent paper presented at the 9th Annual Conference of the American Psychosocial Oncology Society reported that men, who experience frustration and embarrassment about erectile dysfunction after prostate cancer surgery, might be poor adherers to treatment. The authors further describe the potential implementation of ACT to increase their levels of adherence; a clinical randomised control trial is currently ongoing.(436) Because similar feelings of embarrassment and shame of using the CPAP machine were voiced in this sample, the development of a CPAP specific intervention including elements of ACT might be useful.

One participant also mentioned the value of the interview in reducing the resistance to treatment. The possibility of simply expressing one's thought might be as beneficial as other successful psychological interventions. Creative strategies of how this could be cost-effectively introduced to clinical care might be worth investigating. For example, simply asking the individual to keep a diary (written or audio) during the early phases of treatment might present itself as an inexpensive strategy for guided patient discovery and may contribute to improvements in treatment acceptance and adherence.

3.5.2 Integration of CPAP into life

The titration night gave individuals a first experience of what it would mean to integrate CPAP into their life. Some individuals had a negative experience of this overnight study and although there were no explicit references to the potential impact on subsequent adherence, this relationship has been reported by both qualitative(275) and quantitative studies.(276-278)

Following machine purchase, individuals described integrating CPAP into their nightly routine and at a more general level, into their personal life. This process has been outlined in other qualitative studies.(416, 417, 419, 420)

The integration at the nightly level involved habit forming, slowly getting used to the machine and mask and some generated specific techniques to facilitate this process (gradual increase of time on machine, anchoring to existing routines) Adherence is akin to any behaviour change that needs to be practised in order for the action to become habitual and largely an unconscious process. Smith et al. based their music and habit-forming intervention on Triandis Theory of Behaviour, which claims adherence is largely enforced by knowledge of the consequence of not following the recommendations, and practicing the behaviour until it becomes habitual.(437)They randomised 97 patients with OSA to either a music and habit-forming intervention or placebo control (instructions on daily vitamin intake). The active component included an audio-tape supported with relaxing music, instructing the patient on how to fit the mask comfortably and then introduced some relaxation strategies. The intervention also included the provision of details on the consequences of untreated OSA, CPAP reminder placards and a 4-week diary. The placebo intervention was matched for time, effort and healthcare contact. The active intervention was related to increased adherence, 89% were considered adherent (≥4hrs on 64% of nights) in the intervention group compared to only 55% of the placebo controls after 1 month, however there were no differences at 3 and 6 months. These results indicate that this type of habit-forming technique might be useful in the short-term, and might benefit from supplementation with other interventions to increase long-term CPAP use. A possible limitation of this study was that the intervention included a number of components: relaxation that might have led to improvements in sleep, audio instructions and reminder placards that would have increased habit forming and keeping a diary to note

their experiences, might have made individuals more aware of their situation and actively contemplating reasons for and against adherence.

Integration of CPAP into the nightly routine impacted on some individual's usual nightly habits; especially interesting was the perceived disruption to sleep onset latency. Although some were aware that they were comparing the status quo to a time of increased sleepiness, these responses indicate an increased attention towards sleep, which might further develop into a chronic problem(135, 136) The prevalence of co-morbid insomnia in this population is still very much under researched, despite its high rates of comorbidity of approx. 40-50%(343, 344) and effects on adherence.(334)

Side effects that were commonly reported in the literature (such as skin irritations, air leakages and marks from the straps) were also described in this sample and this was not confined to those who were not adherent, similar to what is reported in other studies, one needs to use the machine in order to experience adverse events related to CPAP.(322) It seems in this sample individuals did experience side effects, however with the appropriate psychological and social resources available to them were able to persevere despite these experienced difficulties, similar to the interviewed samples in other qualitative studies.(419, 421) These results indicate a possible interaction between presence of side effects and psychosocial factors, which might explain the conflicting results found in the literature.

The integration of CPAP also occurred at the level of the individual's personal life, most specifically the potential impact on social relationships was noted as a concern. Noise of the machine could disrupt partner's sleep and intimacy difficulties were often mentioned. This might have been more prominent in those whose bed-partner was more easily aroused by external stimuli. One study indicated in a 2-week period post CPAP initiation, the number of nights of co-sleeping was positively and strongly related to mean hours of CPAP use on days used (r=.74) in 10 male OSA patients.(336) History of co-sleeping (prior to CPAP) with wife was also related to adherence, with only 43 % of those sleeping separate from their wives using CPAP more than 4 hrs per night, compared to 74% of those co-sleeping. One can only hypothesise that this is a result of fear of arousability from sleep. Unfortunately the two-week assessment period is too limiting to determine whether an actual increase in co-sleeping events predicts increased CPAP use (with increased co-sleeping potentially being a

marker of improvements in wife's sleep quality). Larger longitudinal studies are needed to understand the direction of these relationships.

Interestingly in this sample, individuals conceptualised integration of CPAP also in terms of the future, and through the course of the study a certain cognitive shift became apparent. The initial understanding for some individuals was that CPAP was a "cure", with more energy they would be able to exercise and eventually come off the machine. This was especially pertinent for younger participants, who described a fear of having to use this machine for the rest of their lives. There is some evidence, albeit weak, of a positive relationship between increasing age and CPAP adherence.(195, 199, 277) These results support the hypothesis outlined in chapter 2, an interaction between age and psychological factors, potentially younger individuals will have different thoughts about/attitudes to the machine than their older counterparts. Towards the end of the study, however individuals saw CPAP as a crutch for a **chronic** problem and at the end of the three months, most participants accepted that this was a treatment for life, and had fully integrated the treatment into their life.

3.5.3 Motivators and Resources

Individuals described how their improvements in daily and nightly symptoms represented a significant motivator for CPAP adherence, a finding that has been reported in a number of other qualitative studies.(275, 415, 417, 419, 420, 424, 425) Interestingly, one qualitative study reported that individuals described initial difficulties with interpreting subtle improvements and often used their families' feedback and experimentation with non-adherence as evidence for improvements.(421) Changes in OSA symptoms (e.g. decreased sleepiness, increased daytime functioning) post treatment is a strong predictor of CPAP adherence in quantitative studies(179, 181, 182, 193, 195, 200, 325), and this relationship presents itself in a dose-dependent manner.(1, 57, 59) Interestingly, Crawford and colleagues reported, based on results from merging three randomised placebo controlled trials, approximately 29% of the symptomatic improvement is a result of an expectation of benefit.(1) This indicates that the relationships reported between adherence and symptomatic improvement, might not only be based on physiological, but also psychological effects.

This study also reported a strong correlation between CPAP and placebo use, which might be indicative of an "adherent-patient characteristic" (see chapter 5). This trait might extend to concurrent adherence to pharmacological treatments, which might be partly contributing to the benefits observed in these individuals, however this is subject of conflicting reports.(438-440). This potential trait might also be representative of an individual's willingness and determination to 'comply' with treatment. This was certainly described by some individuals in this chapter as a resourse for adherence. Additional resources included an active coping style and active engagement with the therapy (e.g. experimenting with the machine). The former has been reported in one of the qualitative studies(419) and one quantitative study has highlighted the link between active coping measured prior to treatment initiation and increased CPAP adherence at week 1.(333) Active engagement (e.g. experimenting with the machine) seemed to be an important facilitator of adherence. This is similar to the theme that emerged in Dickerson and Kennedy's qualitative study of individuals taking part in a CPAP support group.(416) Experts in the group encouraged empowerment and ownership, being involved in problem solving. These findings fit well with recent developments towards the notion of adherence/concordance versus compliance, the former encouraging much more patient invovlement in care.(173, 441)

Importantly though, some expressed a reason for adherent behaviour was because the doctor had "told them so". This potentially highlights the need for tailoring approaches to individual patients' preferences. Pioneering work from Christensen and colleagues has been described in chapter 2, which reports an interaction between the complexity of treatment for renal failure and patient preference for level of involvement in the treatment is a useful predictor of patient adherence.(407)

OSA not only impacts on the actual patient, but also on the surrounding individuals. Thus, in this sample, the impact of OSA on the spouse/family and potential improvements as a result of CPAP use, were noted as significant motivators for adherence. Nocturnal symptoms like snoring and apneas may contribute to a reduction in the bed partner's sleep quality. Daytime symptoms (e.g. irritability, sleepiness, low motivation) might negatively impact on social relationships/marital satisfaction and potential (and actual) reduction of this impact might motivate individuals to use (and continue using) the machine. This

has been reported by both qualitative(415, 420, 423) and quantitative studies.(307, 336) For example, in a parallel trial comparing 44 individuals treated with CPAP with 25 on conservative treatment (weight loss, sleeping posture and reduced alcohol intake), McFadyen reported both increase in partner reported marital satisfaction (r=.3) and decrease in disagreement frequency in the relationship (r=-.4), were related to increased CPAP use.(307)The direction of this relationship remains unclear from these correlational results: however it supports the possibility of a complex relationship between use and marital functioning. However, because baseline assessments of these measures were not significantly related to CPAP use at 3 months, it might be the potential/actual *improvement* in marital satisfaction that results in increased motivation for CPAP adherence. In Cartwright et al.'s study outlined above,(336) co-sleeping was associated with improved adherence. It was not possible to determine improvements in co-sleeping habits, however one could hypothesise that this indicates that the impact on the wife's sleep quality (lowered arousability) might be related to adherence and subsequent spousal support might be moderated by the impact on the partner's sleep.

Support provided a vital external resource for the participants in the current study. Support from the spouse/family, other CPAP users and medical care team was described as facilitating CPAP use, and is in concordance with reports from the current qualitative studies outlined in the introduction.(415-417, 419-421, 423, 424) This corroborates the findings in qualitative studies that have highlighted the association between improved adherence and spousal support and/or relationship quality(298, 338) and augmented medical support in some(345, 437), but not all studies, which might have been a result of sophisticated 'treatment as usual' groups(442) or small sample sizes.(186, 443).

A few individuals however reported well-meant support from either a family member or the physician was interpreted as pressurising, which might have negative effects on adherence, as reported by Baron and colleagues, who found a negative correlation between spousal pressure and CPAP adherence at 3 months.(338) Additionally, individuals referred by their spouse are reported to present with lower adherence rates compared to self-referred patients.(345) An empathic, non-judgmental and supportive therapist style is associated with increased motivation to change(444) and CPAP interventions that support this style (motivational interviewing/enhancement strategies) have been successfully

explored as techniques for improving CPAP adherence.(353-355, 358) It might therefore be useful to encourage the spouse (and physician) to be more supportive and empathic and less confrontational.

Some participants described being dissatisfied with the educational support provided by the health care team. The education session included information about OSA and the consequences of this sleep problem, knowledge some individuals had obtained on their own account prior to the overnight titration study. These individuals noted a desire for more detailed instructions and guidance on how to implement the treatment to the CPAP machine. This is similar to Smith et al.'s qualitative study, where although individuals most frequently desired information about OSA, this theme was closely followed by need to learn about how to successfully integrate CPAP into everyday life.(417) Goal setting has been successfully established as a strategy for increasing performance in organisational settings, and setting goals that are specific and challenging result in superior performance compared to a lacking of or vague goals e.g. 'do your best'.(445) This might be applicable for CPAP users, where specific goals that are explicit about how to facilitate adherence e.g. 'speak to partner about how CPAP might impact on your nightly routine' and difficult, but achievable goals 'use the machine for the full duration of your sleep time', might lead to increased adherence compared to 'try to use the machine for at least 4 hours a night to avoid potential health consequences of OSA'. Goal setting techniques have recently been adapted for improving health behaviour change, e.g. adherence to lower back pain rehabilitation program,(446) dietary behaviour change,(447) and exercise(448) to name a few.

3.5.4 Summary of themes and their interrelations

Figure 6 highlights each of the three themes, their associated subthemes as well as their interrelations. The themes "conflict", "integration" and "motivators and resources" are represented across two categories: 1) internal and external to the patient and 2) more global and nocturnal issues. The internal conflict between wanting to feel normal during the day and feeling abnormal with the mask at night, and trying to resolve this conflict is characteristic of the process of early CPAP acceptance. However these feelings may persist long into treatment for some individuals and influence adherence. Thus the theme

conflict is presented in the top left corner with amber coloured arrows crossing both dichotomies of each categories; representing conflict as an overarching theme that underlies the other two. Once the CPAP machine has been accepted, the individual will evaluate how this intervention is best integrated into both their personal life and their nightly routine. In order to accomplish this task, both internal and external motivators and resources are utilised. The ease of integration might then encourage increased motivation and the utilisation of certain resources and the red arrows drawn between these two themes indicate this reciprocal relationship.

3.5.5 Limitations and Future Directions

The major limitation of this study was the small sample size with only six having completed all three interviews. Although, no new emerging themes were present in the baseline interviews of the individuals who did not complete all three interviews (n=5), it is difficult to gauge whether the content of these themes was different, as described in Ayow et al.'s study(424). The analysis of the six completers was the most appropriate approach to ensure homogeneity of the sample, as required for qualitative studies.(422)

One further limitation is the largely male sample, which is representative of most studies of OSA, because of the approximate two-fold increase of sleep related breathing prevalence in males compared to women.(10) Any comparisons across gender are not possible here, thus future qualitative studies might benefit from including larger female samples to understand further potential gender differences in CPAP experience.

For reasons of convenience and feasibility, interviews were completed at the sleep centre; it was familiar to patients and most interviews coincided with the titration/follow-up appointment. However, this might have increased the demand characteristics, so although individuals were briefed before the interview that all response would remain anonymous, they might have linked the interviewer with the care provision team and feared any transmission of information, especially about non-adherence. A related consideration is the type of individual who is most inclined to take part in (qualitative) research. These individuals might be more outspoken and willing to discuss their experience with treatment. Future qualitative studies might benefit from the use of audio

diaries(see 139) completed at home on a nightly basis, which could reduce the impact of these limitations on the emerging themes.

As with all qualitative studies, the aim is not to generalise, but to draw conclusions about the particular group of individuals. As described in the introduction, there are a number of qualitative studies that have been conducted, thus this study adds to the "database" of accounts of individuals undergoing CPAP treatment and supports the commonalities outlined above. CPAP isn't something individuals want to do, is associated with a stigma of being ill, and helpless; integration is difficult, but individuals' find ways to push through that resistance, if the benefits outweigh the costs and they have a good support system. However this study has to an extent highlighted the need for examining an interaction between more biomedical/treatment specific factors and psychosocial profile of the individual as described in chapter 2. Future qualitative and quantitative studies might benefit from further investigations of possible interactions outlined from the results in this chapter: side effects, level of involvement, age and their interaction with various psychosocial factors.

3.6 Summary

In summary, this study reveals that CPAP remains a difficult treatment, which needs specific consideration of how to integrate into nightly routines and personal life, and this process relies on internal and external resources and motivators. CPAP adherence remains a complex issue with possible interacting factors, yet it is suffice to say: *CPAP remains logical, rational, and yet people just don't want to do it*.

Chapter 4- A thematic analysis of patient experience of cognitive behaviour therapy for insomnia (CBT-I)

4.1 Abstract

Despite established evidence for the efficacy of Cognitive Behavioural Therapy as the treatment choice for chronic insomnia, the experience of patients implementing the therapeutic components remains relatively unexplored. Patient adherence forms a large part of this treatment experience and thus, this chapter offers a holistic and qualitative perspective on patient adherence to CBT-I. Semi-structured interviews were conducted with individuals with insomnia, who had completed a cognitive-behavioural programme for the treatment of their sleep problem. A total of eleven individuals took part in this study (female n=8). Using thematic analysis, three themes emerged that portrayed the experience of adhering to CBT-I: 'Making sense of CBT-I', 'Ongoing evaluation of components' and 'Obstacles to implementation'. Each theme was associated with three subthemes that provide a more nuanced and interpretive account of how individuals experience the implementation of cognitive behavioural strategies. These results offer unique insight into the issues these individuals faced during CBT-I, and contribute to our understanding of adherence in this population.

4.2 Introduction

Despite the substantive evidence for the efficacy of cognitive behaviour therapies for insomnia only few have explored the patient's experience of implementing CBT-I components. The relatively limited exploration might be a result of the nature of the intervention itself.(206) Chapter 1 has outlined quantitative studies that have identified variables that might be predictive of adherence to this multi-component therapy as well as individual treatment strategies. Although these studies add to our knowledge of what influences adherence, they provide little information about the treatment perceptions, which can be valuable, as they have been associated with treatment choice(449, 450) and more importantly, patient adherence.(451-453)

The quantitative exploration of treatment perception and experiences has been examined in sleep research. In fact, a study conducted as part of this post-graduate research project, but not included in this thesis, found that, in a head-to-head study comparing CPAP therapy with a mandibular advancement device (MAD), individuals diagnosed with sleep apnoea rated MAD as more convenient. Additionally, treatment acceptability predicted CPAP adherence, but not use of the mandibular device. However, the sample size in this study was small (n=34) and treatment perceptions were measured after a 2-week acclimatisation period, rather than at baseline.(454) This study was presented at the Associated Professional Sleep Societies Conference in Minneapolis 2011 in poster format and is included in Appendix 5.

In the insomnia literature, treatment perception has been established as an important feature at various stages of treatment. Certain beliefs prior to any implementation might have implications for subsequent behaviour.(120, 230, 455, 456) Stinson and colleagues showed that endorsement of certain beliefs, such as poor efficacy and attractiveness ratings, or lack of awareness of possible options, might act as barriers for treatment seeking behaviour(455) Unfortunately, this was a retrospective study and differential beliefs towards types of treatment were not obtained. Other studies have examined pre-treatment perceptions towards different psycho-behavioural options(456) and psychological versus pharmacological options(120, 230)

In 431 individuals with primary insomnia, pre-treatment perception (suitability, appropriateness, effectiveness and willingness to adhere) was related to treatment choice (sleep hygiene, stimulus control, sleep restriction or multi-component treatment with these three interventions).(456) Results in this study indicated that individuals preferred multi-component options and stimulus control, and preference was influenced by the perception of the acceptability of each treatment. In one of the earlier studies on pre-treatment perception, Morin and colleagues presented 39 individuals with chronic insomnia and their significant others (n=32) with descriptions of both behavioural and pharmacological treatment options.(120) Results indicated both groups rated the former as more acceptable than medication in all areas assessed, apart from perceived effectiveness in alleviating problems in the short term. These finding were largely replicated in a similar study of 43 individuals with primary insomnia. Pre-treatment perceptions were also found to be a strong predictor of subsequent attrition rates, but not treatment outcome in 37 participants who completed a CBT-I program.(230) The studies so far have not differentiated between insomnia phenotypes; however this might be important factor involved in treatment perception and experience. A recent study expanded this line of investigation to group of individuals diagnosed with psychophysiological and idiopathic insomnia.(457) Replicating the studies above, descriptions of psychological treatments (acceptance-based and behavioural intervention) were preferred over a pharmacological option by both groups; however the idiopathic group rated acceptance treatment more favourable in treating their sleep problem.

These studies highlight the importance of pre-treatment perceptions; a number of studies has also investigated treatment appraisal after completion of the intervention.(209, 230) In the previously mentioned study, Vincent and colleagues asked patient to report on their satisfaction (usefulness and likability) of different components of CBT-I. Perhaps not surprisingly, sleep restriction was rated the least, and sleep hygiene the most favourable. However, strong correlations were reported between satisfaction with sleep restriction and improvements in sleep (r=.6) and quality of life (r=.66-.85); this relationship might have been a marker of increased adherence in those who favoured this option. These results stood in contrast to earlier reports where 65 individuals

with sleep maintenance insomnia, sleep hygiene was rated the least likable compared to the meditation and stimulus control options.(209)

Despite evidence for long-term effectiveness of CBT-I,(458-461) only one study has examined individual's long-term use of CBT-I components.(206) Here, patients were asked to complete a questionnaire in regards to their use of 10 core components. Mirroring some of the results above, relaxation strategies were most often used (70% of respondents), whilst stimulus control/sleep restriction and cognitive restructuring were only used by about 40% of the respondents. The lowest reported use was for imagery techniques at 19%. Largest improvements were however witnessed in those implementing stimulus control and sleep restriction.

Overall, these studies allude to the importance of pre- and post-treatment perception for treatment choice and adherence to CBT-I. In summary, it is evident that individuals find psychological treatments more acceptable than pharmacological options. Conclusions on comparative perceptions across different CBT-I components are less definitive, however there seems to be a tendency for sleep restriction to be least liked amongst these individuals, but its implementation is predictive of clinical improvements. One possible, but very speculative interpretation of these results is, that after a long history of having tried or been offered a variety of pharmacological options, individuals present to the behavioural sleep medicine expert extremely motivated to embark on the psychological route. However, once introduced to the individual components- some of which are behaviourally quite demanding and require significant behaviour change- motivation might be reduced for the following reasons: sleep restriction and stimulus control (1) appear at least initially, to be counter-intuitive, even paradoxical to the individual's goal of increasing their total sleep time(139) (2) they inflict slight sleep deprivation as homeostatic pressure for sleep is increased(462), which can lead to amplified daytime sleepiness and low mood(139) and (3) they include curtailing certain behaviours (e.g. spending longer in bed) that have become engrained during the chronicity of their condition. This might explain why in the some studies these behavioural components are least preferred and implemented. For a more nuanced insight into these perceptions especially in regard to treatment adherence, there is a need to move beyond the top-down approach, where individuals are asked to indicate a level of agreement with researcher imposed statements, and

corroborate the present knowledge with patient-generated perceptions especially on treatment adherence using qualitative, bottom up methodologies. Furthermore, this will allow to expand on these quantitative accounts of treatment perceptions, which constitutes a small part of the overall treatment experience.

To date, only two such studies have been conducted.(139, 463) One study embarked on a systematic investigation of patient's perceptions and experiences of implementing a sleep restriction programme(139). Using both validated self-report measures and qualitative accounts of the treatment through audio diaries twice a day and interviews, Kyle and colleagues were able to map, in detail, the patient's general experience of sleep restriction therapy (SRT) in 18 individuals with primary insomnia. Explicit evaluation of treatment adherence was obtained through the diary entry approx. 30 minutes after awakening and questions on implementation at the 4-week post-treatment interview. Of the total sample, 14 individuals completed the qualitative component of this study. Adjustment issues and difficulties implementing the program was frequently recorded in both the audio-diaries and interviews. Particularly problematic was the impact sleep restriction had on daytime functioning and dealing with the sleepiness. Individuals were concerned over the impact on functioning the following day and boredom or loneliness that were induced by the extended wake period. Certain coping strategies were employed to counteract these effects (e.g., engage in wakefulness promoting activities), however giving in to these effects (e.g. by napping) led to unwanted effects (increased sleep onset latency) that spurred on continued adherence. This study highlights the complexity and effort associated with the implementation of sleep restriction, and provides further information of patient's experience of adhering to this behavioural intervention. Although useful in contributing to the understanding the potential mechanistic principles of SRT, the presentation of SRT in isolation is not routine clinical practice. The common presentation of sleep restriction alongside other behavioural and cognitive components might drastically alter the experience of individual components, with patients on the one side allowed to "pick and choose", but also the combination of components may exacerbate the effects of individual treatment. For example, the repeated getting up after 15 minutes of wakefulness with a restricted window of about 5 ½ hours could be challenging.

Currently only one study has investigated the experience of completing to a multi-component package CBT-I from a qualitative perspective.(463) Open-response feedback forms with questions regarding their treatment experience were completed by 43 of the total 52 participants with chronic insomnia after completion of the 7-week CBT-I program. The responses were subsequently coded into themes, indicating that about half of the participants reported the completion of CBT-I in a group format and sharing the insomnia experience with others was most helpful. Although the strength of this study was the qualitative analysis of patient generated responses, there are some shortcomings of this study. Firstly, it did not provide an in-depth qualitative analysis of the patient experience and secondly, there was no explicit focus on treatment adherence.

The undeniable benefit of such qualitative exploration is to provide rich insight into patient's perceptions and experience of implementing components, which are not fully obtainable through the previously outlined quantitative approaches. This aim of this chapter is thus to expand on these investigations and present an in-depth qualitative exploration of patient's experience to CBT-I. With a particular focus on adherence, this study investigates some of the complex issues individuals are faced with when implementing components of CBT-I, which will not only advance our understanding of adherence to this treatment, but potentially provide information about treatment delivery and interventions to facilitate implementation.

4.3 Methods

4.3.1 Participants

Individuals with insomnia, who had partaken in research studies that included a manualised form of CBT-I (either as part of the core research protocol, or as a good-will gesture for participating in other research), were approached. To reduce the variability in treatment delivery, only those who had undergone manualised CBT-I as described elsewhere(207, 464, 465) were included in the study. This format encompasses five sessions covering sleep hygiene/education, stimulus control, sleep restriction and cognitive restructuring components. A full description of this CBT-I program can be found elsewhere.(207, 464)

Further inclusion criteria were:

1. Participants had to be aged between 18-65 years

2. have completed treatment within the last 24 months (this was decided to minimise effects of recall bias; however all eligible participants had completed the treatment within the last 1-4 months)
3. have had a diagnosis, prior to CBT-I initiation, of primary insomnia according to the research diagnostic criteria [RDC(92), see Appendix 1]. (As all participants had completed the CBT-I intervention as part of other research projects, this information was obtained from the research data base)
4. fluent in the English language

Those with co-morbid psychiatric or medical disorders were excluded unless there was a temporal and causal separation from their primary sleep concern, in concordance with the research diagnostic criteria.(92) The presence of a co-morbid sleep disorder (e.g. sleep apnoea, restless legs, parasomnia) was a further exclusion criterion. Individuals who were also using other forms of treatment for their insomnia (e.g. pharmacotherapy) were not excluded (as above, this information was obtained from the information on the research database).

Twelve eligible individuals were identified and approached, of which one was unable to attend the interview because of travel distance to the research centre. This is an adequate sample size for this type of study.(422)

4.3.2 Procedure

Ethical approval was obtained from two committees NHS Greater Glasgow and Clyde and Sydney Local Health District Ethics Review (RPAH Zone) Committees to conduct this study at two separate sites (University of Glasgow Sleep Centre, Glasgow, UK and Woolcock Institute of Medical Research, Sydney, Australia). Information sheet and consent form are presented in Appendices 6 and 2 respectively.

Once contacted and their consent obtained, participants were invited to the research centres for a face-to-face interview. All interviews were conducted by the lead researcher during 2009-2010, and questions and prompts were pre-determined (see Appendix 4). This enabled the interviewer to entirely focus on the responses, permitting each individual to speak freely and with as little guidance from the interviewer as possible. Thus the direction of the interview was malleable depending on issues that were generated by the respondent.

These types of semi-structured formats enable the exploration of the individual's experience, thoughts and beliefs in rich detail.(422)

After explaining the study rationale (see interview schedule in Appendix 4) the participant was reminded that the interview would be tape recorded. Suggested topics covered in the earlier parts of the interview were mainly descriptive in nature e.g. 'history of the sleep problem', allowing the interviewer to establish rapport with the participant. Once the interviewer felt the participant was comfortable, more sensitive topics were discussed 'feelings when treatment was suggested' or 'impact CBT-I has on every day life'. Table 8 highlights the topics covered in the interviews. Duration of the interviews was between 40-90 minutes. Each interview was transcribed verbatim and subsequently analysed using thematic analysis.

Table 8: Topics covered in semi-structured interviews

The experience of insomnia and participation in CBT-I
Factors that influenced their decision to adhere/not adhere
What factors they believed influenced adherence to other treatments, and relating this back to their experience with CBT-I
What they would have changed in the process to facilitate adherence

4.3.3 Data preparation and analysis

Interview data were analysed using thematic analysis (Braun and Clarke 2006) as outlined in chapter 3. The results in this chapter are presented slightly differently from chapter 3. Because individuals were only interviewed at one time point, quotes are presented within the text forming a narrative account of the individuals' experience of adhering to CBT-I.

4.4 Results

4.4.1 Patient Characteristics

The group of 11 participants was relatively homogeneous. The majority were female (73%), and well educated. For the 10 participants who provided this information, the mean years of education was 16 (SD=3). All individuals gave details about their ethnicity; the largest proportion described themselves as White (British/Scottish, Australian or Canadian, n=10, 91%) and followed by Asian (n=1, 9%). Individuals considered themselves to have had their sleep

problem for an average of 10 years (SD=8). Approximately half were married or had a live in Partner (55%), whilst others were single, divorced or separated (45%).

Table 9 presents the characteristics of each participant.

Table 9: Participant Characteristics

Participant Alias	Age (Yrs)	Education (Yrs)	Marital Status	Insomnia Duration (Yrs)	Post CBT-I Insomnia Severity Index (/28)	CBT–I Group
Belinda	**63**	**10**	**Divorced, living alone**	**20**	**16**	**1**
Ben	**45**	**17**	**Separated, living alone**	**7**	**8**	**1**
Sally	**58**	**17**	**Live-in Partner**	**13**	**-**	**2**
Mike	**44**	**19.5**	**Live-in Partner**	**10**	**12**	**2**
Jonathan	**47**	**16**	**Married**	**6**	**11**	**2**
Caroline	**58**	**11**	**Married**	**4**	**12**	**3**
Madeline	**51**	**-**	**Single**	**6**	**18**	**3**
Abigail	**22**	**16**	**Single, living with friends**	**.7**	**3**	**4**
Polly	**49**	**16**	**Divorced, Living alone**	**9**	**4**	**4**
Rachel	**43**	**16**	**Married**	**30**	**-**	**4**
Karen	**48**	**19**	**Live-in Partner**	**4.5**	**2**	**4**

4.4.2 Qualitative Results

Three themes emerged from the thematic analysis of the interviews: “Making sense of CBT-I”, “Ongoing evaluation of components” and “Obstacles to implementation’. A summary of these and their associated subthemes are detailed in Table 10 below. In the following, each theme is presented with the respective subthemes and patient quotes embedded in the text to feature the experience of patient adherence to CBT-I in a narrative style.

4.4.2.1 Making sense of CBT-I

Individuals described their initial impressions at the initiation of treatment. Very few had concrete conceptualisation of what CBT-I actually really was, and individuals were willing to “give it a go” as CBT-I presented itself as the last resort. CBT-I was also conceptualised in reference to the meaning of their insomnia.

4.4.2.1.1 What is CBT-I really?

At initiation of CBT-I, it was uncommon for participants to have a tangible understanding of the treatment. Very few had further investigated this type of

therapy and many even described entering treatment with a 'blank canvas'. Individuals, like Belinda, were mostly concerned with the prospect of improving their situation: *'I don't know that I really had any expectations, I think I just hoped that, you know, I would learn things and at the end of the day, put them into action and would learn how to sleep a bit better' [Belinda, lines 139-141].*

Apart from identifying CBT-I as "talking therapy", individuals were unsure of the content of the therapy:

> *'I thought it might have been like therapy of some sort, like talking about your problems, how you got them, what we can do about them. I wasn't sure what, in what format though.* […] *I thought it was a short thing, I thought "oh I will just go for some tips […] and I can take them away for me and try and work them you know".'* (Polly, l. 93-99)

> *'well I wasn't sort of sure, I guess, what we were going to go into, like I said, I have never looked into doing anything for my sleep before* […] *so I didn't know… I assumed…I guess now it makes sense that there are things like sleep diaries and thing, but I never considered I might be filling one of those out, so not in terms of content, not a lot, particularly.'* (Abigail, l. 136-139)

The simple distinction from the familiar pharmacological options seemed to be a motivator to take part in something individuals had little knowledge about:

> *'I didn't really know what to expect I think it was just going in there almost, like with a sort of blank canvas, open minded, willing to try and take on… I wouldn't have been willing to take on any medication as such, ah, as I said, I've, I just don't like taking medication basically.'* (Ben, l. 205-210)

> *'when you approached me and it was non-drug, I thought "why not give it a go… can't lose anything".'* (Rachel, l. 85-86)

> *'there was no harm in trying, it was not like I was going to you know, like if you are taking drugs get addicted to it, and not have anything, not having a result out of it, so I wasn't loosing anything, there was only something to gain. So, that was motivation for me.'* (Rachel, l. 390-393)

4.4.2.1.2 CBT-I as the last resort

In the interviews it emerged that, for the majority, CBT-I was their last hope for improvement; having tried various other treatment options (pharmacotherapy,

hypnotherapy and herbal remedies) individuals were willing to engage in this treatment that was, as Karen describes, the "*last port of call" [l.154]':*

> *'I either do this, or I…see I didn't see that not sleeping anymore would actually get better, so it was get worse or get better.'* (Karen, l. 154-156)

Individuals described the emotional impact of sleeplessness; feeling "depressed" and "fed up", at the end of their tether, which resulted in a sense of emotional "relief", when the treatment was suggested:

> *'When I got the chance of this, I thought well, I can't go on like this. Not sleeping or sleeping for a week, you know, and I find sometimes [sighs] sometimes, it really gets you down, you feel quite tired and depressed; because you are not sleeping you know.'* (Belinda, l. 54-58)

> *'I was fed up, I couldn't, well you can't say you couldn't go on, I was just like, every day was getting worse and I was fed up and you know I thought I tried all these herbal remedies and they were like not really working, so I thought, well "I will go to the sleep clinic".'* (Madeline, l. 217-220)

> *'I think it was a bit of a relief once I started coming here, wouldn't say I saw immediate benefits, but I think you felt as though, yeah, well there is a way out of this.* [...] *there is a light at the end of the tunnel here.'* (Ben, l.123-129)

CBT-I was seen as the solution- both in terms of leading to improvements in sleep, and in providing an acceptable alternative to pharmacotherapy, a route they were less comfortable in pursuing. The combination of feeling helpless /at the end of their tether, and CBT-I taking shape as an option that wouldn't produce any side effects, or would not be something they were in danger of becoming addicted to, proved a catalyst for engagement in this therapy:

> *'I wanted to see it* [CBT-I] *through, I wanted a positive outcome, I mean and this was a chance like I said, I can't see any other avenues to go down, so if this didn't work, I didn't know what else to do. And I can't…I think I would have ended up taking more pills.'* (Caroline, l. 1098-1100)

> *'You know that everything was worth a try, I mean obviously I hadn't gone the sort of medication route, as it was, which is something I could have discussed with my GP, if I had wanted to. So on some level, I*

have always avoided that, so you know, sort of doing therapy for it, seemed like a good idea.' (Abigail, l. 46-49)

Table 10: Themes and Subthemes that emerged from the interviews using thematic analysis

Themes	Subthemes
Making sense of CBT-I	What is CBT-I really?
	CBT-I as the last resort
	The meaning of insomnia
Ongoing evaluation of components	Evaluation against past, present and future self
	Using what works
	Comparing self to others
Obstacles to implementation	Is the timing right?
	Beliefs about sleep
	Negative/Unwanted Consequences

4.4.2.1.3 The meaning of insomnia

Making sense of CBT-I occurred in light of how these individuals conceptualised their insomnia. Although many described the frustration, helplessness and hopelessness of having insomnia and were ready to embark on the changes outlined in the treatment; some participants described an initial disbelief in the potential effectiveness of CBT-I, based on the meaning insomnia had for them. What emerged in some interviews is that for CBT-I was seen as too simple to deal with their complex, chronic problem. An excerpt of the interview with Rachel highlights how she defined her insomnia as a **chronic** problem- chronic to the point she had accepted it was just the way she slept. This perception influenced how she made sense of CBT-I:

Rachel: I didn't think it would work.

Interviewer: Why was that?

Rachel: When you first called I thought 'oh this might be a waste of time' [laughs]

Interviewer: What was the reason for that?

Rachel: I don't know, maybe because I have had this problem for so long, then I just thought it was the way I slept, or lack of sleep. I just thought

it was me. And I guess you get to a point where you just accept what you have and just get on with it.

(Rachel, 130-140)

Others defined their insomnia in terms of the complexity of their problem. For Karen, CBT-I components seemed 'too simple' for her complex problem, whereas for Ben it was the process of sleeping that was too simple to deserve treatment:

'some of the basic techniques of not turning the lights on at night, of you know winding down before I go to bed, of when I do wake up going "it's not such a big deal and I will cope tomorrow and I will be fine." They sounded too simple to actually work.' (Karen, l. 90-93)

'I suppose I was a bit apprehensive, 'cos [sic] it's something new and you feel as though a bit stupid, really, thinking you know, "how can you not get to sleep?" […] surely you can resolve this yourself without somebody having to tell you.' (Ben, l. 114-119)

Interestingly though, when the attempt was made to implement these techniques, individuals realised how difficult some of the components actually were. Karen, who had initially understood CBT-I as too simple, made this discovery:

'Putting them into practice was actually harder than I had expected. You know the getting up after the 15 minute rule was really hard, and you know a couple of times I have slipped on the waking up at 6 o'clock especially in the mornings.' (Karen, l. 93-96)

In contrast, other's felt CBT-I was a suitable option for their chronic complex sleep problem:

'It [CBT-I] wasn't something synthetic, and the…there were going to be skills, like, you know presumably would you know last a life time, in terms of being helpful. It didn't seem like such a, what would you say, like a band-aid, you know it was more of a long term solution.' (Abigail, l. 67-70)

'So that is how I look on this treatment too. So it's not- it will, it may not help me in the shor…well does help me in the short term, but the long-term is what I am looking for. Yeah, long-term benefits.' (Polly, l. 827-829)

On the one side, insomnia was defined as a debilitating, chronic condition affecting daytime functioning and quality of life; however participants also described an alternative meaning of insomnia. For a small number of interviewees, insomnia was defined as was part of one's identity; an excuse; or even a blessing in disguise, providing the individual with more opportunities during night-time hours, which led to the initial 'trepidation' or resistance towards CBT-I.

> '*Trepidation* [described feeling when treatment was suggested], *because I was thinking "oh gosh", you know "will this work really? "Am I", you know, "am I going to be able to get out of it" because I realise that it was a habit, and I kind of even sometimes looked, not sort of looked forward to being up all night, but I thought, "well I can always do that when I go to bed".'* (Sally, l. 101-104)

> '*it's shedding those things that make you different, and having the badge of being an insomniac, it's like "oh no, shedding that one too".'* (Karen, l. 121-123)

> '*before it was quite easy to say "oh look", you know "leave me alone, I have had a bad night" or "the reason I, I am not thinking straight is because I had a bad night". so there is no more blame, to having a problem with sleep, because it is not there now.'* (Rachel, l. 395-396)

4.4.2.2 Ongoing evaluation of components

It emerged in the interviews that individuals did not simply **comply** with what was outlined in the programme, but experienced a process of evaluation of the components on which they based their subsequent adherence behaviour. Each aspect of CBT-I was continuously evaluated in terms of efficacy: individuals used what worked; what worked for them; and in terms of what worked for others.

4.4.2.2.1 Evaluation against past, current, future self

Once initial impressions had been made and the treatment was established as an option that was in concordance with the way individuals perceived their insomnia, there was a more detailed evaluation of the treatment components in respect to the image they had of themselves. Treatment components were deemed valid when there was concordance with individuals' past, current and future self-image.

Newly imposed routines were compared to how they used to be, or seen as a mechanism to establish the old sleep patterns:

> *'that's what I used to do, so maybe, maybe if I start trying that again, and its really just, re- its sort of like, it reaffirms the "yes, I have been on the wrong track and there is a way back.'* (Sally, l. 460-462)

> *I thought "oh this is great maybe they can come up with some different strategies, or ideas that would help me re-establish my old sleep pattern".'* (Polly l.74-76)

Participants also continuously assessed whether or not there was an alignment between the treatment components and their current habits and preferences. This could either encourage or discourage their continued use of the particular components: Abigail, who described herself as not having '*whole lot of negative thoughts'* found using the cognitive strategies very difficult. Similarly, Mike who disliked *'routine imposed upon'* him sometimes struggled to stick with the sleep restriction programme. For Belinda, Abigail and Madeline, there was congruence between what was required of them and the description of their "self" as someone who '*doesn't lie in bed any length of time' 'ha[s] never been someone to snooze the alarm' or* '*would try anything'* respectively.

Others again identified their present circadian preference or insomnia subtype when evaluating treatment components:

> *'I still am a night person, but I think I realise also there is better things, there is opportunities at the other end of the scale, other end of the day as well. Yeah, it's [sleep restriction] sort of opened my eyes a bit really.'* (Polly, l. 443-446)

> *'my problem doesn't seem to be going to sleep, my problem is staying asleep. So it is difficult to stick to the 15 minute rule you know, when you are lying there at 2 o'clock in the morning and you are feeling shattered, it is difficult, although I have done it.'* (Belinda, l.247-250)

In addition to their image of their past and current self, patients also described their future self and the goals they aimed to achieve in terms of their sleep: *'I still see the challenge to get over that because its not quite where I would want to be, but at least there is like a direction to go.' (Jonathan, l.154-156).* The prospect of reaching these goals provided added motivation to adhere

to the treatment components. For some it was also the image of what they could become, if they failed to initiate change that provided a push to continue:

> *'With older people, like my mum is in a nursing home at the moment, and when I get there, it doesn't matter what time I go there, they are all asleep, a lot of them are napping, I thought "crikey if I fall into that category now, what hope have I got when I am that age" I don't what to get into that.'* (Polly, l.397-400)

4.4.2.2.2 Using what works

Of components that were deemed appropriate for themselves, individuals used what proved effective in improving their sleep patterns. The following quotes reflect how this involved an ongoing process:

> *'It's like "ok I will test that bit out, no that doesn't work, I will test…no that doesn't work, oh that works, ok I will keep doing that one" so yeah for me it's really about trial and error and about what works and doesn't work.'* (Karen, l. 664-667)

> *'I just went through the processes of what you told us, and if one didn't work, I would do the other, and eventually one would work.'* (Rachel, l.200-201)

Some individuals identified the specific components that worked for them. Interestingly, no single component emerged as the most useful and it becomes evident, that these individuals used what worked best for them.

> '*the relaxation tape to help you relax before going to bed … I found that useful you know, I found that did help me get to sleep quicker.'* (Jonathan, l.94-95)

> *'Building up that sleep drive and sticking to a sleep window has been…I felt really good about that, that it works and can be something that one can use.'* (Mike, l.144-145)

> *'the relaxation tape, I tried that, but I couldn't be bothered with it, it just, I think, I thought it was too long winded, so I got frustrated with that, so I didn't like to use that. But I like the fact of having stories, creating yourself a story and the picture in your mind, and setting yourself somewhere else I liked that. And the chanting the name and writing off all your thoughts before you go to sleep, that worked for me.'* (Rachel, l.459-462)

> *'I don't imagine anything, I just don't let my mind wonder, if it starts wondering, I think that is when you start thinking, so its just something that I haven't used, but I don't think I would use it, because the other things are working for me.'* (Caroline, l. 732-734)

There was a dissonance between using what worked and the actual difficulty in adhering to the components. Individuals described almost a cost-benefit evaluation when deciding to follow recommendation. In the majority of cases, like for Madeline, the effectiveness seemed to outweigh any difficulties individuals were having and provided the motivator for continued use.

> *'I don't want to get up and get out of bed, when you don't have to and you know you are going to go back there shortly, so I would rather not, but I mean it does work, you get up and you go have a little wonder and then you go back.'* (Madeline, l.778-781)

Seeing the effects of non-adherence provided a form of validation for the efficacy of components and this increased their motivation to be more rigorous with the treatment routine. Seemingly testing the boundaries is imperative in increasing their awareness of how treatment works and how non-adherence affects outcome.

> *'I woke up at about half past 5, I could have got up then quite easily and went, "no I will just sneak some more sleep in" […], then I felt pretty, not as alert all day, just a bit sort of lethargic, and then that night, I just had so much trouble getting to sleep, […] thinking "I don't want to do that again, see a direct causal relationship between when I get up in the morning and when I go to sleep the night before".' (Karen, l.511-517)*

> *'Saturday night, come bedtime I wasn't tired, I didn't feel tired, […] but that was my bedtime, so I kind of footed about and went to bed, which was a big mistake because I didn't sleep all night.' (Belinda, l.440-445)*

4.4.2.2.3 Comparing self to others

Lastly, patients also evaluated components after comparing their behaviour with others. This could be others in the treatment group, or reports of previous success stories. Seeing how components led to improvements for others in the group or hearing about the effectiveness of CBT-I from the current literature

provided a source of validating the effectiveness of these components and encouraged continued use:

> *'I didn't think the thought blocking… I didn't actually think would be that helpful to be honest. And then one day, I remember I have forgotten her name, who had done the '2' instead of the 'the' and she had said you know that that had been really helpful, so then I thought 'oh I might as well give it a go.'* (Abigail, l. 441-445)

> *'it helps with […] listen to someone who is similar, but approaches it in a different way and going "oh, ok if they are similar to me and then if I approach it their way, it might work for me too".'* (Karen, l. 881-884)

These comparisons also gave hope that the improvements would been seen if they continued to adhere to components:

> *'And I think it was just getting that sort of reassurance; this and this worked for all these different people, there is every chance that it will work for me too.'* (Ben, l. 548-550)

In this process of comparing themselves to others, individuals also recognised dysfunctional behaviour or thought processes, they were also trying to change. As both Sally and Karen describe, this process helped them internalise the underlying treatment rationale to provide validation of the treatment components:

> *'It's like me giving the advice that I don't take myself really, […] and it's a kind of re-affirmation, yeah, when I am trying to say, "well you try this and if you try thi..." it's as if I am trying to convince myself that these things will work and then by explaining to someone else, it helps me get it straight in my head really.'* (Sally, l. 216-220)

> *'I do really like the group thing and hearing how other people going, and hearing myself in other people who aren't going so well, and recognising parts of me, but I think it's not judging them, but it's going "they are still hanging on to that bit, I am too", if you know, and learning that, that reflective thing of "if it is not working for them, why do I think it is going to work for me?".'* (Karen, l. 868-872)

4.4.2.3 Obstacles to implementation

Throughout the CBT-I programme, individuals were faced with certain obstacles that made implementation of the treatment components challenging. Trying to

implement components at the wrong time, remaining dysfunctional beliefs and the presence of unwanted consequences were obstacles described by these individuals:

4.4.2.3.1 Is the timing right?

Optimal behaviour change occurs at the correct time. This could be at the day-to-day level with adherence issues occurring on difficult days, or at the broader contextual/motivational level- individuals have to be ready for change with treatment initiation occurring at the right time in life. Unfortunate timing might then result in practical or motivational obstacles for implementation. Implementing the sleep restriction programme on the weekends or during holidays was particularly challenging for many individuals in this study. Karen describes the reason she found this difficult was because it is "traditional" to stay in bed longer on the weekends. As the following quotes highlight, partners and families were also impacted during these times:

> *'as my partner pointed out, how can you do this during the holidays, why are they meaning you doing [sic] this in the middle of the holidays. So […] given the opportunity, I guess I would do it at a different time' (Mike, l. 534-537)*

> *'you've got weekends and your partner is there, […] you can't just dictate your sort of sleep pattern to yourself, you cannot just be up, I mean you can be, but I generally you not being that selfish, […] so you're sort of you're not really adhering to… in those occasions because you want to spend time together.'* (Ben, l. 368-373)

The sub-theme timing was also considered in terms of the broader contextual level. Both Jonathan and Sally described the timing of CBT-I initiation in terms of their current lifestyle; Jonathan's relatively free schedule facilitated effort in implementing components, whereas Sally's hectic life-style produced obstacles for adherence:

> *'I think I probably had the advantage, that because I am not currently working, it was probably easier for me to organise my time to fit in to the programme, and the steps of the programme, it might have been slightly more tricky for the people who were in work.'* (Jonathan, l.80-83).

> *'it's been […] not a normal period of time sort of thing, so if I had, just like a normal […] if my life had been running normally, then I …it would have been different definitely, but in a way, I might not have been as determined to try and see it through.'* (Sally, l. 673-676)

Individuals also acknowledged that importance of timing in reference to their internal motivation and whether they were ready for change, as Polly denoted, one of the biggest things was *'that we are willing to change, I think, […] that we are actually willing to.'* (l. 936-937) Some individuals described at the start being *'resistive' 'having trepidation'* or thinking the treatment wouldn't work. Karen explains how this prevented her from initially seeking treatment:

> *'I was really resistive [when CBT-I was suggested], because someone had suggested it when I came for the sleep apnoea test here. And I just put it off and put it off and put if off and put it in the too hard basket.'* (Karen, l.86-88)

4.4.2.3.2 Beliefs about sleep

Beliefs about sleep needs provided a further obstacle for implementation. One belief that prevented many from implementing the behavioural components was aspiration to obtain more sleep, or the magical '8 hours':

> *'I couldn't sit up till 11.30 anyway and get up when I have to get up, I find that I just needed that sleep, but then I then I'd be counting up the hours, so that is a long time I do spend in bed sometimes, when you say you only need 8 hours a night sleep, I must need 8 hours a night sleep.'* (Caroline, l. 423-426)

> *'once I sleep [sic] in […], I could have got up at 7, but I just decided that it wasn't going to do me any good and, and so my sleep window ended at 7, I got up at something like 9.'* (Mike, l. 283-286)

Karen even describes this experience of wanting more sleep as *'an old voice'* that felt validated when she did decide to stay in bed past her rising time- *'"see you did get an extra hours sleep".'*:

> *'it's like when you wake up in the morning you have got a vulture sitting on the end of your bed going "glad you are awake, I want to have a*

word with you"' so it's a nag, a little voice in your head all the time.' (Karen, l.359-361)

For others, it was the understanding (or misunderstanding) of the treatment rationale that was an obstacle:

'if you don't fall asleep you know sometimes I find too, if I do get up and I sit, I become more wide awake and that kind of defeats you know you can be sitting there for a long time before you're feeling tired.' (Belinda, l.253-255)

Interestingly, individuals explain how 'getting their head' round the treatment rationale, facilitated the rebuttal of these dysfunctional beliefs. For some this process was about truly understanding the rationale of the components and comprehending the consequences this had on their behaviour. Yet again for others, like Karen it was about re-defining the rationale, to make sense of it on a personal level. Rather than taking it literally, people individualised the treatment components to get their head around them and fully internalise the rationale.

"I really need to get my head round this as well, if I don't feel tired when I should be going to bed, then I just don't go" (Belinda, l. 443-445)

'the effort of physically getting up, I just think "augh, no I will just lie here and I will fall asleep eventually", whereas generally speaking that's not what happened, you do drop off, but I felt as though in those occasions, you weren't getting a proper sleep.' (Ben, l. 335-358)

'Well, it has been working a bit better, because you are going to bed late, you are not going to bed because you are just like 'oh might as well go to bed, I am tired", but you really aren't as tired as you should be, you got [sic] to get that sleepy tired. So that is quite good, that window of sleep, I think and it does make you, I think it makes you sleep a bit more too.' (Madeline, l. 144-148)

'the 15 minute rule, its kind of good, but for me its more about the point at which the frustration sets to start in, that is, and that can happen after 5 minutes, so that for me is the more important part of that bit, because that is when the anxiety and the distress start to happen, so if I get up at that point, and break that thought cycle.' (Karen, l. 762-766)

For Karen it wasn't solely about internalising the rationale, but also internalising sleep and accepting sleep as part of her self, which made the CBT-I experience less fearful:

> *'Well, is it [sleep] something to be conquered? That is the thing, its just, I don't conquer being awake […] but for me sleep actually has to be so normal, that is part of who I am and part of what I do, rather than this almost external entity that has to be gotten better of. […] So to normalise sleep, makes it less of a foe, less of something to be afraid of, yeah I think that works for me.'* (Karen, l. 384-393)

4.4.2.3.3 Negative/Unwanted Consequences

Individuals described difficulties implementing the behavioural components- especially sleep restriction and stimulus control- when these produced unwanted or even unexpected consequences. One of the most frequently noted adverse effect was increase in sleepiness. Despite their willingness to adhere, they found it progressively more difficult to ward off these feelings that emerged during treatment.

> *'I was tired. I struggled with the sleep restriction on the basis, I… it was suggested that I shouldn't be going to bed until about 1 in the morning and I just physically couldn't stay awake until that time.'* (Ben, l. 149-151)

> *'it was really slow mornings then a period of feeling fine and then early evenings starting to get really tired and, having to stick with it and stay up and stay out of bed and so it was challenging.'* (Mike, l.154-157)

> *'I was quite tired most of the nights, like that I had out, I couldn't actually stay out as late as I used to, because I was feeling more tired.'* (Abigail, l. 269-270)

Apart from more physiological consequences, the presences of unwanted practical consequences inhibited implementation of treatment components. Individuals felt that some components impacted on daily activities and especially on others in the household:

> *'sitting up at night, and getting up early in the morning, when you live in a small house, with people, who were a bit sensitive to noise, at*

first I was like "oh, what am I going to do", kind of, you can't turn the telly on, because the noise.' (Karen, l.209-211)

'*then she [partner] was on holiday for a week after and I wanted an alarm going off at 7".' (Mike, l. 187)*

In order to overcome these side effects, participants described the need to develop certain coping strategies. For many participants implementing change was more difficult that expected and side effects like tiredness or boredom had to be counteracted by these strategies. For example countering sleepiness by staying active, changing habits like sitting on the couch, or finding activities that would encourage rising in the morning, facilitated adherence. Likewise, individuals had to develop strategies to prevent the more practical unwanted side effects that disrupted others in the household:

'In the evening, I had to stay physically active to be able to… you know, if I'd sit down for a long time, I would really get sleepy, so I had to stay physically active to…, in order to stick to the programme.' (Mike, l.176-178)

'Ah if I don't sit in the lounge, I think I am just too exhausted, I have to sit somewhere, because I have been going and going, *if I sit at the dinning table, which is a nice firm chair, then I definitely stay awake.'* (Polly, l.348-350)

And my partner did suggest that I do the laundry and other things and I said 'no' the motivation to get up at 6 would just disappear instantly of course. You know to do the housework, so, yeah it had to be something that I actually enjoy doing. (Karen, l.224-226)

"How to set the alarm and finding something that wouldn't wake the whole house up- just an ipod under the pillow with a low volume, it woke me up, but not my partner" (Karen, l. 258-260)

4.5 Discussion

In 2006, a review on the use of psychological and behavioural treatments for insomnia concluded that effectiveness studies were lacking.(466) Behavioural adherence is a significant component of effectiveness of any treatment, yet our understanding of adherence to CBT for insomnia is equally narrow. This study aimed to explore the experience of individuals undergoing CBT-I in order to shed light on this phenomenon. Interviews were conducted to investigate how people

follow recommendations to overcome their insomnia and the salient issues that influence their behaviour.

Three superordinate themes were extracted from the analysis that reflected the overall experience of implementing CBT-I components: “making sense of CBT-I”, “ongoing evaluation of components” and “obstacles to implementation”. Figure 7 provides a summary of the themes and their respective subthemes. How these themes are related is explained further below.

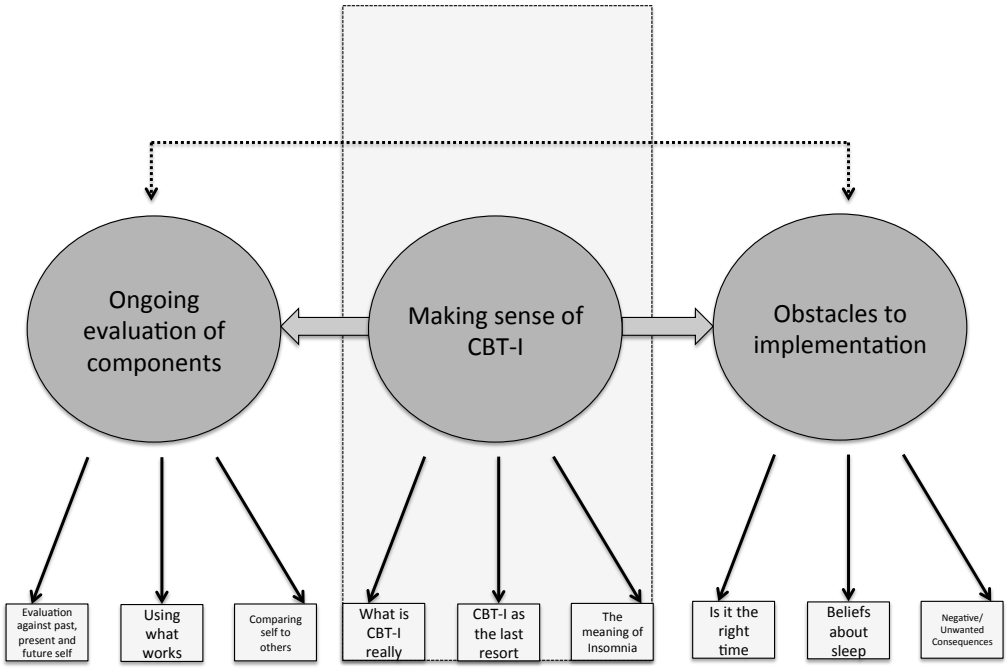


Figure 7: Themes and Subthemes and their Interrelations

4.5.1 Making sense of CBT-I

This theme reflected how individuals obtained a global sense of what CBT-I was. At the start of treatment, these individuals had little conceptualisations of the content of CBT-I, having entered the treatment with no prior knowledge or research into this type of intervention. They were willing to engage in treatment, potentially because it offered a solution that was different from all other options previously experimented with (pharmacotherapy, herbal remedies etc.) Little awareness of sleep and available treatments, as reported in our sample, is potentially a function of sleep having little presence in public health. A study conducted in the UK by Stores and Crawford found that sleep is acknowledged merely 5 minutes in undergraduate medical student’s curriculum(467) With general practitioner appointments, certainly in the UK and Australia, limited to around 15 minutes, the opportunity to speak about sleep

during consultations is even further reduced. The consideration of increasing awareness and availability of CBT-I has been addressed elsewhere(121) and there is evidence to show that lack of awareness can act as a barrier to seeking treatment for insomnia,(455) however the concrete relationship between awareness/knowledge and subsequent adherence to CBT-I remains unclear in this population. In the majority of respiratory sleep clinics, sleep education plays a large role in introducing continuous positive airway pressure (CPAP) to individuals with obstructive sleep apnoea (OSA) however educational interventions have not been associated with or produce only small improvements in CPAP use, see chapter 1. Further research might benefit from investigating how psychoeducation in CBT-I is related to adherence to other components.

In this sample, individuals described a debilitating history of trying to manage their sleep problem prior to the start of CBT-I. It was common for individuals to have tried a variety of alternative options prior to seeking professional help with their sleep, which has previously been reported in the quantitative literature(107) also in qualitative studies.(139, 468) Many described their insomnia as debilitating condition, which corroborates previous qualitative investigations of the emotional and psychological effect of chronic poor sleep(468-470). This sense of being at the end of their tether, coupled with the ineffectiveness of previous pharmacological and self-help attempts, led to a sense of relief when CBT-I was suggested. CBT-I was described as 'light at the end of the tunnel', however this also translated into this treatment being identified as their last resort. If taking part in this opportunity did not prove effective, there was no other option. It is difficult to draw conclusion on how exactly this might have impacted on individual's adherence, however one possibility is that the effect of CBT-I being scarce might have encouraged individual's motivation to initiate and adhere to treatment (perhaps even more so than if these treatments were readily available). This suggestion is speculative; however the scarcity effect is well established in the social psychology/marketing literature.(471) This is not to say that CBT-I should remain scarce and recent efforts to develop strategies to improve the availability of CBT-I(121) are laudable, however therapist might strategically draw on this in their efforts to improve adherence.

Lastly, individuals made sense of CBT-I in light of how they defined their insomnia. For some their sleep problem was defined in terms of its chronicity or

complexity and the anticipated level of effectiveness or suitability of the treatment was contrasted with this view. There is a large body of evidence that has revealed the relationship between illness perceptions and health behaviour.(472) Leventhal's self-regulatory model suggests that an individual processes information about the illness and forms a cognitive representation thereof on the basis of 5 dimensions: identity (what are the symptoms, labelling the illness), cure/control (curability and controllability of the illness), cause, consequences and timeline (potential duration of the illness).(473) Whether these types of perceptions are related to adherence to CBT-I is relatively under-investigated. One study however found that the perception of insomnia as a trivial condition that does not to be addressed within a therapeutic framework was an obstacle for seeking treatment.(455) Using the Illness Perception Questionnaire (derived from Leventhal's model), Morgan and colleagues(232) found no relationship between perceptions about the curability/controllability of the sleep problem and adherence (personal communication). However, this study did not investigate the other illness perceptions and had used the crude measure of attendance as a marker of adherence.

4.5.2 Ongoing evaluation of components

Patients described how they continuously evaluated their self, the components and others throughout treatment. This theme portrays not only how dynamic the process of adherence is, but that for these patients, treatment implementation is not merely about complying/not complying, but an active decision making process to adhere or not to adhere.

Firstly, patients critically evaluated the components against their self in past, present and future form. Past and future images of self reflected either a healthy sleep pattern they were hoping to return to/achieve or the downhill spiral into poorer sleep habits they recognised from pre-treatment times and hoping to avoid. The image of their present self took on many different facets, from general habits to more sleep related perceptions of the self (type of insomnia: onset versus maintenance, or circadian preference). A dissonance between the treatment and the image of their self led to a poorer evaluation of the treatment components, which were subsequently not implemented. If patients are choosing the aspects that overlap with the image of their self, there might be value in matching treatment components across assessed criteria. The

few efforts to individualise treatment have, however been unfruitful.(474, 475) Unfortunately, these treatments were tailored according to poorly established methods of subtyping (e.g. using questionnaires to match cognitive intrusion or physical tension with cognitive therapy or relaxation therapy respectively). Efforts to match treatment against relatively more robust diagnostic distinctions of subtypes of insomnia (paradoxical, idiopathic, psychophysiological insomnia) could be more fertile for the possibility of individualised treatment. For example,(457) reported that individuals with idiopathic insomnia rated acceptance-based therapies as more preferable than their psychophysiological counterparts.

Patients made further inferences about the individual treatment components – selected for implementation were components that were effective and easy for them to use. There was a selective decision to use some components, whereas others were discarded, rated as less suitable or effective. Treatment preference and attribution has been given relatively little attention in the insomnia literature. Although some studies have reported that psychological/behavioural treatments are preferred over pharmacological options when assessed prior to treatment(120, 230, 456), there are only a few studies that have attempted to establish which of the actual treatment components are rated most acceptable and thus most often used.(206, 209, 230) It is difficult to draw conclusions from these studies-with different research designs- about individuals differential preferences for each aspect of CBT-I. This study certainly was not able to gauge which components were most favoured by individuals, and it was also not the intention of this study to investigate in detail perceptions about each component, however it seems that individuals used the elements they perceived as most effective for them.

The experience of occasional non-adherence was used as an important marker in determining what worked and this inadvertently increased their willingness to comply. Failure to implement medical recommendations is inherent to most treatments, even behaviour as simple as pill taking is associated with poor adherence rates.(169) This experience provided patients with a strategy to validate the treatment: non-adherence lets them re-discover how their sleep had been before treatment initiation. To avoid individuals abandoning treatment entirely, preparation for relapse should be an integral part of CBT-I.(see 207) Currently, motivational interviewing/ enhancement

therapy (MI/ME), which acknowledges relapse as an important part of change, has been proven useful for problem behaviours like substance abuse.(476) Some studies have provided preliminary evidence for the effectiveness of MI/ME in improving CPAP use.(354, 355, 358)

Lastly, witnessing the effectiveness of components in other group members or hearing of previous success stories, led to a re-evaluation of the suitability of treatment components for individuals. Bandura's social cognitive model states that people form knowledge about their environment from observing and modelling other's behaviour.(340) It has yet to be explored whether this phenomenon plays a role in adherence to CBT-I. Group administration of CBT-I is well-established and leads to clinically meaningful improvements in sleep outcome variables(140) even when compared to the one-on-one delivery option.(477, 478) Despite the consensus that group therapy might increase adherence to treatment components,(477-479) and one qualitative study reported that individuals valued meeting others like them during group CBT-I(463), there has been no systematic investigation of adherence across these different delivery modalities.

Similarly, the incorporation of previous treatment completers has not been considered, but the effectiveness of including such strategies in a CBT-I program to improve adherence could be investigated. A recent study in OSA reported that the incorporation of short 15-minute video of current CPAP users in a cognitive behavioural intervention, displaying both treatment success and failure, provided an increase in average nightly machine use of 2.9 hours compared to treatment as usual.(351)

4.5.3 Obstacles to implementation

Despite the extensive literature on the effectiveness CBT-I, few studies have studied potential obstacles for adherence to this treatment. In this study despite having made sense of CBT-I and evaluated which treatment components were most suited to their needs, individuals identified certain features of the process that created obstruction to implementation. The practical, motivational and external obstacles overlap to an extent with what has been established in the literature: increased initial wake periods caused by stimulus control or sleep restriction may lead to boredom and annoyance(139, 208), or increased sleepiness.(139, 218)

In this study timing was identified as one of these obstacles, with individuals describing weekends or holidays as the most difficult times to adhere to the sleep scheduling practices. The social norm of 'sleeping in' or additional social activities on weekends and holidays might make adhering to the get-up time increasingly difficult. To date the self-report scale that have been used to measure adherence to the behavioural components has not included separate items for week versus weekdays.(208, 214) However, considering that this subtheme emerged in both this and Kyle's qualitative investigation(139) of the experience of sleep restriction therapy, the differentiation of weekday and weekend in future adherence scales might be fruitful. Of note, this type of adherence self-report measure has been developed at the University of Glasgow research group and is considered in more detail in chapter 6. Participants in this sample also identified how an important facilitator for treatment adherence was the implementation of CBT-I at the right time in life, both from a practical, but also motivational perspective. The motivation or readiness to change has been extensively studied in the framework of the transtheoretical model. This theory describes behaviour change can only occur when the individual is at a stage ready for change (see chapter 6 for a detailed outline of the transtheoretical model). The consensus in the literature is that individuals with insomnia are ready for change once treatment is sought; however when faced with the behavioural components and the prospect of reducing the time they spend in bed, which might seem counter-intuitive, motivation might be altered in some individuals. Certain dysfunctional beliefs that remain despite cognitive restructuring elements of CBT-I might play a role in this reduction in readiness to change.

Beliefs about sleep (especially sleep needs) emerged in this study as a further obstacle for treatment implementation. If the dysfunctional belief of needing 8 hours of sleep is still heavily endorsed, removing oneself from bed in accordance with sleep restriction and stimulus control will be challenging. Their goal of increasing their sleep might seem counter-intuitive with what is asked of them. This experience was also highlighted in Kyle's qualitative study(139). The themes termed 'this is a sleep restriction programme' and 'adherence and adjustment' highlighted participants' frustration with limiting (at least initially) their sleep opportunity, when their goal is paradoxically to increase this. A successful restructuring of individual's thought processes prior to

implementation of the behavioural components might be an interesting avenue to pursue. Unfortunately, to date there has been no systematic investigation of the order of cognitive and behavioural elements of CBT-I. Most packages will provide the behavioural prior to cognitive components. However, this and the previous subtheme (timing at the motivational level) suggest it might be useful, at least in some individuals, to offer the cognitive components first. Randomising individuals to either pathways, or matching the pathway according to individuals' beliefs and readiness to change could be useful, as has been shown for alcohol dependence.(480)

In terms of dysfunctional beliefs, research has indicated that these are subject to change across the treatment of CBT-I; additionally, improvements in certain cognitions- such as needing 8 hours of sleep- were associated with improvements in outcome(146, 481). Whether this process is important for adherence should be subject to further quantitative and qualitative investigations.

Finally, the experience of negative/unwanted consequences emerged as an obstacle to implementation. The more practical impacts on the family or daily activities were seen as frustrating side effects of adherence, however even more potent were the physiological consequences of acutely increasing their homeostatic drive for sleep. Mounting feelings of sleepiness- an experience individuals with insomnia are often unfamiliar with- left individuals challenged in adhering to the behavioural components of CBT-I. This phenomenon of adverse effects has largely been neglected in the research of psychological-/behavioural treatments for insomnia.(123, 139) Kyle et al. indicated that in the acute stages of a sleep restriction programme side effects -especially extreme sleepiness- emerged in both the qualitative and quantitative measures of this study.(139) As described in chapter 1, interventions that target these obstacles e.g. introduction of modafinil, might have a positive effect on adherence.(see 218)

Individuals in this study identified coping strategies, such as finding activities or eliminating sleepiness inducing environments, to overcome the physiological consequences and found solutions to reduce the impact of the more practical consequences. Evidently these individuals are successfully developing active problem solving techniques to overcome these obstacles. Both the presence of an active coping style and increasing self-management has been associated with increased adherence to CPAP,(333, 482) whether improving

these strategies along side the implementation of CBT-I components remains to be investigated.

4.5.4 Summary of themes and their interrelations

Figure 7 summarises the themes and their respective subthemes. Making sense of CBT-I at the global level is core to the process of implementing the components, as indicated by the framed box in Figure 7. Establishing the meaning of this treatment is fundamental and with this in place, individuals begin to focus their concerns on the individual component. This incorporates both the evaluation of each component against several criteria and also how certain obstacles may complicate the implementation. These themes "ongoing evaluation" and "obstacles" might inform each other (as indicated by the dashed arrow in Figure 7). For example, beliefs about sleep as an obstacle might cloud the evaluation of the component's effectiveness. Likewise, something valued as effective might facilitate the continued implementation despite marked obstacles. However, this was not entirely clear from the data and remains a speculative interpretation.

4.5.5 Limitations and Future Directions

It is important to evaluate this study in light of its limitations. The aim of this- akin to all qualitative studies- was not to produce results that are generalisable to the population, but to describe the experience of this particular sample. There is a need for further qualitative and quantitative studies to explore the themes highlighted here, for us to make generalisable statements about the experience of CBT-I and important factors that influence adherence. One limitation of this study is it sampled participants from both the UK and Australia. Samples in qualitative studies are generally comprised of a homogenous group. Recruiting participants from only one cultural background might have generated different qualitative data; on the other hand, themes emerged in this qualitative investigation, are reflective of experiences across both settings, relatively uninhibited by effects of different cultures and health systems. Only individuals who had completed the same manualised form of CBT-I were included, which minimised effects of using a culturally heterogeneous group. There were no apparent differences in themes emerging across these two sites.

One further limitation is the largely female, well-educated sample of individuals with primary insomnia, selected from previous research studies. Although this is quite representative of research samples within the insomnia literature, it will be imperative to investigate experiences of a real-world clinical sample that in addition to insomnia, might be dealing with a co-morbid sleep disorder like sleep apnoea, or other physical (e.g. pain) or psychological conditions (depression)- exclusion criteria in the present study. Some of these conditions have been associated with decreased adherence to CBT-I(215) and qualitative investigations in these subgroups might provide further insight into adherence to CBT-I. Furthermore, the experiences of a largely motivated, health literate research participant might differ gravely from the journey of someone who is presenting to their GP in their first treatment-seeking attempt. Interestingly though, even this motivated group of research participants, who largely responded to treatment, found CBT-I challenging.

This qualitative investigation relied largely on the recall of experiences in these individuals. All participants were interviewed between 1-4 months after completing the treatment protocol, thus recall effects might have biased retrospective accounts of the experience of implementing components. Additionally, the retrospective nature of the study resulted in the exclusion of treatment drop-outs, this might have led to the omission of important experiences related to the early phases of CBT-I unique to this group. Future studies might want to follow individuals through the entire CBT-I process to obtain a more detailed account of individuals experience in real time. Techniques such as audio diaries, with which individuals can record their experiences on a daily basis, has successfully been used in a previous study involving insomnia population(139) and might provide a valuable resource for this endeavour.

Lastly, for reasons of feasibility, the interviewer in this study was also the therapist or shadowed the therapist in all CBT-I groups. This might have led to the interviews being biased by demand characteristics. However, there was no evidence of this bias in the results as presented. .

4.6 Summary

This present study is the first to extensively explore the patient's experience of Cognitive Behavioural Therapy for insomnia. This has provided us with valuable information about how, within this group of patients, adherence is a very dynamic process: patients continuously evaluated CBT-I and the components. This started as early as at treatment initiation, and continued throughout treatment. Patients engage in various thought processes in determining which treatment components to implement and do not simply comply. Additionally, this study provides further evidence that the implementation of some of the behavioural components is far from simple and this needs to be considered in future studies trying to understanding the treatment experience and important factors that influence behavioural adherence to CBT-I.

Chapter 5- The effect of continuous positive airway pressure usage on sleepiness in obstructive sleep apnoea: real effects or expectation of benefit?

5.1 Abstract

Rationale: Placebo responses are complex psychobiological phenomena and often involve patient expectation of benefit. With continuous positive airway pressure (CPAP) treatment of obstructive sleep apnoea, greater hours of CPAP use are associated with reduced sleepiness. However, these open label studies have not controlled for patient expectation of benefit derived from their knowledge of their hours of device use. **Objectives:** To investigate the relative effectiveness of the use of real or placebo CPAP on daytime sleepiness. **Methods:** Patient-level meta-analysis combining data on sleepiness measured by the Epworth Sleepiness Scale from three randomised placebo-controlled crossover trials (RCTs). Mixed model analysis of variance was used to quantify the effects of real vs. placebo-device treatment, usage, their interaction, and regression to the mean. **Measurements and Main Results:** Duration of real and placebo-CPAP use was correlated within patients ($r=0.53$, $p<0.001$). High use of real CPAP reduced sleepiness more than high use of placebo (difference 3.0 points; 1.7 to 4.3, $p<0.0001$) and more than low use of real CPAP (difference 3.3; 1.9 to 4.7, $p<0.0001$). High use of placebo was superior to low use of placebo (difference 1.5; 0.1 to 2.8, $p=0.03$). Twenty-nine percent of the effect of high usage of CPAP (4.2 points; 3.3, 5.1) was explained by the expectation of benefit effect associated with high use of placebo (1.2; 0.2, 2.3).**Conclusions**: A clinically significant proportion of the effectiveness of high CPAP use in reducing sleepiness is probably caused by patient expectation of benefit.

5.2 Introduction

Poor compliance with medical treatments is a major barrier to clinical effectiveness for many chronic conditions.(483, 484) Recent studies suggest that placebo effects may involve crucial psychobiological factors influencing

treatment effectiveness and clinical practice.(485) Placebo treatments are not necessarily inert; one principal component is the expectation of future benefit responses following administration of a placebo.(485, 486) Patients who accept treatment are expecting that it will help them. These observations have primarily been made in a range of pharmaceutical and other therapeutic interventions. However, the availability of real and placebo continuous positive airway pressure (CPAP) for obstructive sleep apnoea (OSA) provides an almost unique opportunity to examine the relationships between treatment usage, expectation of benefit, relief of symptoms and the placebo effect including expectation of benefit.

Unblinded cohort studies have shown a dose-response relationship where greater usage of CPAP was associated with better symptomatic outcomes for patients.(57-59, 487) However, because those were open-label studies patients were largely aware of how many hours a night they had been using CPAP. Therefore it is possible that some of the symptomatic benefit associated with greater use might have arisen from an expectation of benefit.

Randomised placebo CPAP-controlled crossover trials provide a method to determine whether the symptomatic response to higher use of CPAP is related to a real effect or expectation of benefit from their high use. Given the complexity of human psychobiology, cross-over trials offer a better comparison as patients act very well as their own controls, because usage of real and placebo CPAP seems to be highly correlated.(488) Placebo control via an almost identical sham device also helps to quantify the size of the expectation of benefit effect associated with using CPAP therapy. We combined 3 crossover trials(488-490) in an individual patient-level meta-analysis in order to quantify the relative effects of real and placebo CPAP compliance on sleepiness measured using the Epworth Sleepiness Scale.

5.3 Methods

5.3.1 Participants

We combined data from the 91 patients who completed one of the crossover trials. Detailed descriptions of the study designs, patient's characteristics, and primary outcome findings of both studies can be found in the original

publications.(488-490) Table 11 briefly describes the patients included in this analysis from these studies.

Table 11: Patient Characteristics across the three trials

	Trial 1 (Marshall et al 2005)	Trial 2 (Coughlin et al 2006)	Trial 3 (Phillips et al 2011)
Gender (Female)	7/29 (24% female)	0/34 (0% female)	3/28 (11% female)
Age (Median Years)	50.6 (range 25-67)	49.0 (SD 8.3)	48.8 (range 25-72)
Apnoea Hypopnoea Index	21.6 (SD 7.5)	39.7 (SD 13.8)	38.68 (SD 24.04)
Body Mass Index (kg/m^2)	31.5 (SD 6.0)	36.1 (SD 7.6)	31.7 (SD 4.1)
Baseline Epworth Score	12.5 (SD 4.1)	13.8 (SD 4.9)	10.3 (SD 4.8)
Prescribed CPAP Pressure (cmH_2O)	7 (range 5-10)	10.0 (IQR 8-10)	11 (range 7-18.5)

5.3.2 Study Design and Procedure

Trials were combined as they had congruent study designs with all patients receiving in random order both standard individually titrated real CPAP and placebo CPAP (sometimes called 'sham CPAP' and hereafter simply described here as placebo). The placebo was identical, in all trials to the CPAP machine in terms of noise, mask temperature, mask humidity, and airflow through the exhalation port. To create a sub-therapeutic treatment, the placebo machine was set to 8 cm H_2O, yet delivered <1.0 cm H_2O pressure. The datasets were merged and we analysed the effects of real and placebo treatment and the effects of greater use of both these devices on sleepiness as measured by the Epworth Sleepiness Scale (ESS).(5) The ESS was the only outcome in common among the trials.

5.3.3 Data Preparation and Statistical Analyses

Statistical analyses were performed using SAS (v. 9.2; SAS Institute, Inc., Cary, North Carolina, USA) and SPSS for Windows (v. 17; SPSS, Inc., Chicago, Illinois,

USA). Continuous variables were presented as mean ± standard deviation (SD) or 95% confidence intervals (CI) and p-values of <0.05 were considered statistically significant. We considered the interaction of compliance-by-treatment to be significant when p<0.1 because this was the primary screening method for our hypothesis– the specific group-by-group analyses arising from such an interaction were then judged by the p<0.05 criteria.

We used mixed model analyses of variance (using variance components structure of covariance) to quantify the effects of usage (High vs. Low cut at 4 hours/night) and the interaction between treatment and compliance. These models also included the effects of treatment (CPAP or placebo) and regression to the mean (the baseline severity in ESS). A priori we specifically tested the following comparisons: 1, the superiority of high use of CPAP to high placebo use: 2, the superiority of high CPAP use to low CPAP use and 3, the superiority of high placebo use to low placebo use (all using p<0.05 as the critical threshold level). Inter-trial and inter-individual variability were classified as random effects; all previous variables (treatment, compliance, regression to the mean, and the interaction between treatment and compliance) were fixed effects. We also estimated the proportion of improvement associated with high use in clinical practice that is probably attributable to expectation of benefit by dividing the effect associated with high placebo use by the effect associated with high CPAP use. Effect sizes were calculated by dividing mean effects by the standard deviation of the Epworth Sleepiness Scale, which is very often around 4 points in both clinical and population samples. Small effect sizes are between 0.20 and 0.50, medium 0.50 and 0.80, and large effect sizes .0.80.(491)

These main analyses were conducted using compliance as a *dichotomous* variable based on compliance cut at the standard 4 hours/night; in a second model we investigated the relationship between outcome and compliance as a *continuous* variable using the mixed model analysis described above. In sensitivity analyses we tested different dichotomous cut-points for use and whether the order the treatment was given may have influenced our final conclusions by using an order and an order by treatment interaction in our final model. We also examined the correlation between CPAP and placebo use.

5.4 Results

5.4.1 Correlation between placebo and real CPAP compliance:

Real and placebo device usage was correlated (r^2 = 0.53, p<0.001; see Figure 8). Individuals who used placebo more than 4hrs/ night were also very likely to be high CPAP users with only 3 of those patients having compliance below 4 hours when on CPAP (see Figure 8).

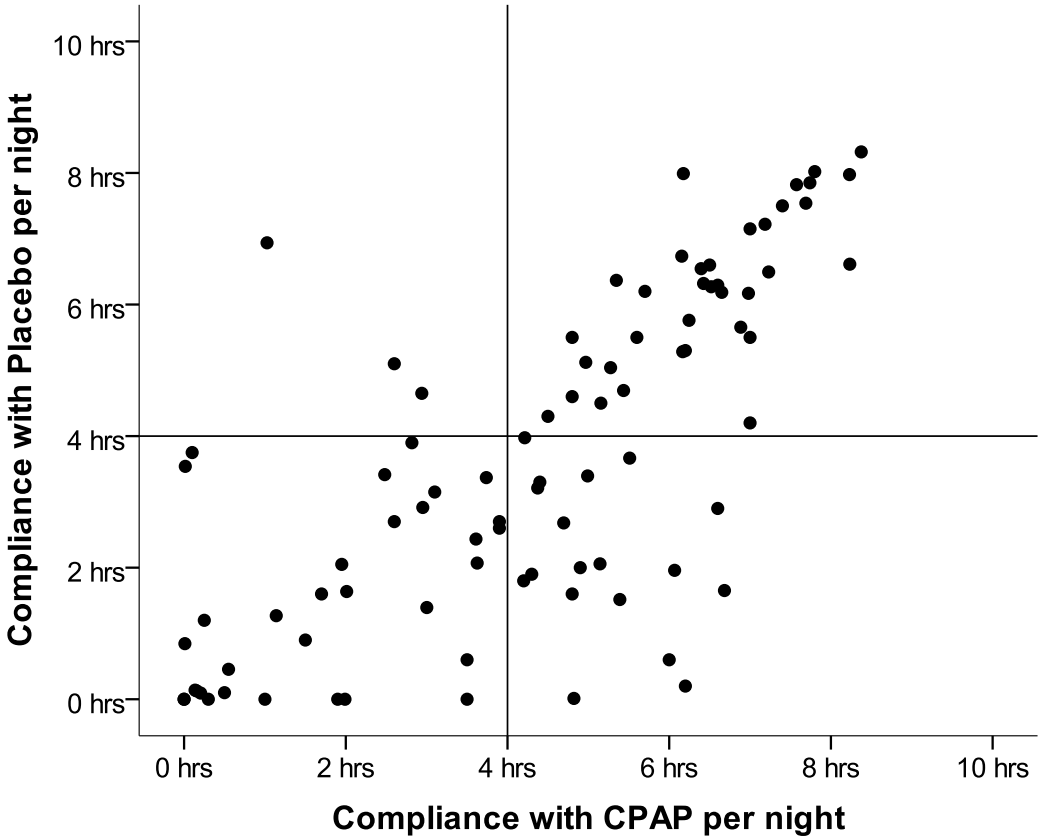


Figure 8: Correlation between real and placebo use
Patient use of treatment is stable between a real CPAP device and a placebo version of the same device (r^2=0.53, p<0.001). X and Y axes are in units of average hours per night of use.

5.4.2 Epworth Sleepiness Scale (ESS)

High use was associated with superior improvement in the ESS across both treatments (mean difference =2.2 points; 95% CI=1.0 to 3.3, p<0.001; effect size (ES) based on a SD of 4 points =0.55). Real CPAP improved sleepiness more than placebo irrespective of usage (2.1; 1.1 to 3.0, p<0.001; ES=0.53). The interaction between hours of usage and type of treatment (real or placebo) was

significant (p=0.056), using a significance threshold level of 0.1 for investigating the specific comparisons of interest. The effect on ESS of high use of real CPAP was greater than higher usage of placebo CPAP (difference=3.0 points; 1.7 to 4.3, p<0.001; ES=0.75) and low use of CPAP (3.3; 1.9 to 4.7, p<0.0001; ES=0.83). Additionally, high placebo use was better than low placebo use (1.3; 0.1 to 2.8, p=0.03; ES=0.33). These effects were not influenced by order or order by treatment interaction. These results are presented in Table 12 and Figure 9.

Table 12: Effects of high and low CPAP and placebo use on Sleepiness

	High vs. Low Use [Mean (95%CI), p-value]	CPAP vs. Placebo [Mean (95%CI), p-value]	Interaction Treat*use [p-value]	High CPAP vs. High Placebo [Mean (95%CI), p-value]	High Placebo vs. Low Placebo [Mean (95%CI), p-value]
ESS (24 points)	2.2 (1.0-3.3), p<0.001	2.1 (1.1-3.0), p<0.001	p=0.056	3.0 (1.7-4.3), p<0.001	1.4 (0.1-2.8), p=0.03

Data are presented as Mean Points of improvement from baseline, (95% CI), p-value. This table presents the effects of compliance, treatment and the interaction between treatment and compliance. The compliance effect tests whether high use is better than low use. The treatment effect tests whether CPAP was better than placebo and the interaction tests whether the effect of high use differs between CPAP and placebo. CPAP=Continuous Positive Airway Pressure; ESS=Epworth Sleepiness Scale; CI=Confidence Intervals

High use of CPAP was associated with a 4.2 point reduction in Epworth (95% CI = 3.3, 5.1 points, p<0.0001) and high use of placebo was associated with a 1.2 point reduction in Epworth (0.2, 2.3 points, p=0.03). As such, we estimate about 29% of the improvement in Epworth scores associated with high CPAP compliance seen in clinical practice is probably attributable to expectation of benefit in patients who are high users of CPAP (i.e. 1.2 points on high use placebo divided by 4.2 points on high use CPAP).

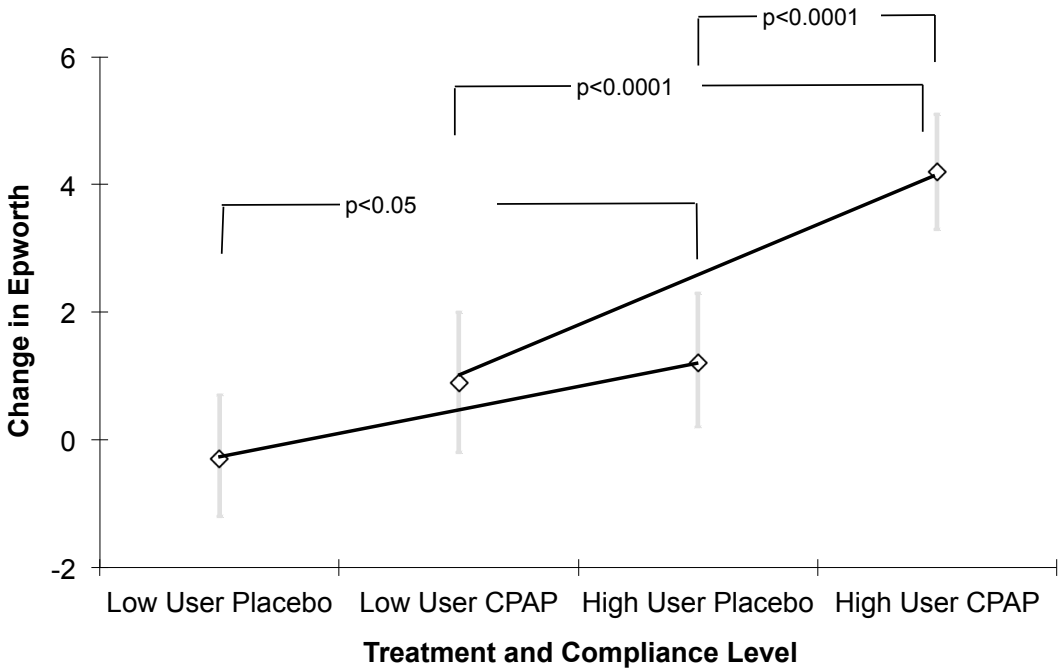


Figure 9: Effects of different treatment type and use on improvements measured by the Epworth Sleepiness Scale

(y axis is in points of improvement from baseline in the 24 point Epworth Sleepiness Scale). Diamonds indicate the estimated mean effects (bars the 95% CI). High use (>4 hrs/night) of either machine confers greater benefits but the symptomatic benefits accruing to high users are greater when using real CPAP than placebo CPAP. CPAP=Continuous Positive Airway Pressure.

We also investigated usage as a continuous linear variable. In these models, both treatment and usage were significantly related to Epworth improvement in the expected direction (mean estimate=2.1 points (95% CI 1.2, 2.9) and 0.6 points per hour, 95% CI 0.3 0.9, respectively, both p<0.0001). In contrast to the findings in the dichotomous model, there was no significant interaction between treatment type and usage duration (p=0.54). In other words, the effect of duration of use, measured as a continuous linear variable, on ESS did not significantly differ between CPAP and placebo devices.

In sensitivity analyses we analysed the cut points for high and low use using 5, 6.5 and 7.5 hours/night. The combination of these analyses plus plotting raw data and fitting curves to it indicated the likely existence of no additional benefit beyond about 5.5 hours, but with significant uncertainty about the exact location of the asymptote (See Appendix 7).

5.5 Discussion

Using data from 3 randomised placebo-CPAP controlled crossover trials, we estimate that 29% of the reduction in sleepiness seen in patients with OSA who use CPAP over 4 hours a night is probably caused by an expectation of benefit. The expectation of benefit is one of the components of the placebo effect and is caused by patients inferring benefit from treatments they choose to use. Conversely, patients who do not use treatments do not expect to feel better. Because clinical interaction is such a complex psychobiological phenomena, these effects are best quantified in randomised cross-over trials where patients act as their own controls and tend to use the active treatment (CPAP) about as much as they use the placebo.

Previous open label cohort studies(57-59) and parallel design clinical trials(492, 493) have shown that there is a dose-dependent association between greater use of CPAP and better patients outcomes.(370) Instead, we analysed three placebo-CPAP controlled randomised crossover trials in order to separate real treatment use effects to symptom improvement related to this placebo-like expectation of benefit within each patient (see Figure 9 and Table 12). As expected real CPAP had a beneficial effect over placebo as did high use of either device compared to low use. Fortunately, not all of the benefits of high use were driven by expectation of benefit as patients using CPAP more than 4 hours a night had greater reduction in sleepiness than patients using placebo more than 4 hours/night. We infer expectation of benefit effects because patients using placebo for more than 4 hours a night exhibited greater reduction in sleepiness than patients who used placebo less than 4 hours a night, despite this device having no physiological effect on OSA. These high users of placebo were also 94% likely to be high users of CPAP (46 out of 49, see Figure 8) indicating the existence of a compliant patient phenotype who will use the device regardless of its ability to control OSA. Conversely, there were also patients who used placebo CPAP less than normal CPAP (see the lower right quadrant of Figure 8). This may be further evidence that patients who feel benefit from CPAP treatment tend to use it more.(59)

The main effect we report here cannot be observed in clinical practice because patients in routine clinical care are not given placebo CPAP therapy.

Additionally, placebo devices are not commonly employed in cross-over trials due to a fear of unblinding,(494) so the number of clinical trials available for this sort of analysis is limited.

The uncertainty about whether the dose effect is linear, stepped or asymptotic led us to treat compliance as both a dichotomous (the widely used 4hrs/night threshold value) and a continuous hours of use variable.(57) When compliance is treated as a continuous linear variable in these same models both treatment with CPAP and high usage confer significant benefits. However, the non-significant interaction term in that linear model indicates that the use effects of CPAP and placebo do not differ in a way that is consistent with a linear-dose response. In sensitivity analyses we dichotomised hours-of-use data at different cut-points (5, 6.5 and 7.5 hours) and additionally visually plotted the raw data (see supplemental figures). These raised questions about the linearity of the relationship between compliance and symptomatic improvement. Thus we can also confirm previous reports that after controlling for placebo effects the association between compliance and better patient outcome is weaker than expected(59) and may not be linear(57) as benefit ceases to accrue at an asymptote of about 5.5-6.5 hours per night.

Figure 8 shows patients using either the real or placebo device in similar amounts. Whether this ‘high-use’ trait might generalise between CPAP and pharmacological compliance is the subject of conflicting reports.(438-440) Additionally, the possibility of modifying psychological characteristics of non-compliance with CPAP treatment should continue to be the subject of clinical trials (e.g. (351) and ACTRN:12606000065594). The characterisation of patients into ‘high’ and ‘low’ users is defensible because CPAP compliance is often bimodally distributed (see Figure 8 and the relative paucity of data points centred around 4 hours compared to 0-2 hours and 6 hours). However this classification may still be flawed as recent investigations of both pharmacological treatment of epilepsy and CPAP indicate there may be more than 2 compliance phenotypes.(205, 495)

The use of crossover trials with placebo control offers only a partial solution to the problem that patients are aware of (unblinded) to their ‘dose’ of treatment. A trial where patients are randomised to 2, 4, 6, or 8 hours/night of effective treatment might offer superior data to what we are currently able to present here. Our approach of using each patient with their highly

correlated(488) use of CPAP and placebo to act as their own control offers an advance on previous analyses. However, such highly selected and motivated patients may not be generalisable to normal clinical populations. Data from both clinical trials and unselected cohorts provide useful clinically applicable information when employed together. We were also limited by the availability of only one subjective outcome variable and by the limited numbers of patients (n=91) available from these technically challenging clinical trials. Two of the trials did not have 2nd arm baseline measurements(489, 490) and one did not have a washout period(490) which might have affected our results. The Australian-based study[10] also had lower baseline ESS scores than would be expected for a symptomatic clinical sample and this may have made detecting treatment and use differences harder because of a possible floor effect. One of the implications of this is that in clinical practice with patients who have very high ESS scores the dose-effect may be stronger than we report here and the dose-benefit may extend well beyond the asymptote we may have detected. We may have had more accurate treatment estimates if we had used some measure of percentage of sleep with CPAP rather than the more crude hours per night- however we lacked a good objective measure of sleep duration. Nevertheless, these rare data provide opportunity for understanding aspects of CPAP compliance that have not yet previously been investigated.

Longstanding efforts to improve compliance to CPAP by employing technological solutions aimed at reducing pressure may be questionable as Figure 8 also suggests compliance has little relationship to pressure. This also explains why the two most recent meta-analyses of pressure modification approaches to improving PAP compliance show little or no compliance benefit and no symptomatic benefit.(177, 245) In this context it is not surprising that the largest effect of any treatment intended to increase use of CPAP therapy tested in a randomised trial has been cognitive behavioural therapy.(351)

5.6 Summary

This patient-level meta-analysis of 3 randomised placebo-CPAP-controlled crossover trials for sleep apnoea patients confirms that high use of CPAP provides greater sleepiness reduction benefits than high use of a placebo device. Interventions that improve CPAP use from low levels (i.e. below 4 hours/night) are likely to cause real benefits for patients.

Chapter 6- The Stage of Change Scale for Insomnia (SOCSI) - A new scale to monitor stage of change during sleep restriction therapy for insomnia

'Patterns of behavio[u]r are not usually created, modified, or stopped in a single moment in time or with a single flick of a switch.'

DiClemente 2006(496, p.25)

6.1 Abstract

A number of patient reported-questionnaires based on the Transtheoretical Stage of Change (SOC) Model have been developed to identify adherence issues in other sleep populations (e.g. CPAP users). However rigorous psychometric testing of these questionnaires is usually lacking. Additionally, no such scale exists to predict adherence to behavioural treatments for insomnia. This study set out to develop a measure to assess individuals' stage of change during a sleep restriction programme for their insomnia. The stage of change scale specific for insomnia (SOCSI) using cognitive interviewing techniques (Study 1) was firstly developed and face validity assessed. Interview responses from individuals who had completed CBT-I treatment (n=13) indicated good face and content validity; participants indicated no missing responses. Some amendments to the scale were recommended by participants and adopted in the final version of the scale. Further psychometric evaluation of the scale considered the validity in predicting adherence to a sleep restriction programme (Study 2). The SOCSI was administered to twenty-seven individuals with primary insomnia who completed the 4-week intervention. Adherence was measured using a self-report questionnaire, sleep diaries and actiwatches, however only data from the latter were correlated with significant sleep-related improvements. Stage of Change was skewed with the majority scoring in the action/maintenance stage at all points throughout the treatment and 3-month follow-up. Stage of Change status and self-efficacy ratings were highly reliable when re-assessed after a 10-minute distractor task. Those who scored in the action stage had better rates of concurrent adherence, motivation and self-efficacy (concurrent validity) and subsequent adherence throughout the 4-week intervention (predictive validity)

period than those in the pre-action stages (contemplation/preparation). Sleep improvements and impaired energy/motivation were most often reported as the pros and cons for change respectively although the majority indicated no disadvantages with the therapy. Behavioural strategies (e.g. self-liberation) were most often reported to help implement change compared to cognitive strategies (consciousness raising). Although the results need to be interpreted in light of the study limitations, documenting stage of change at early periods during therapy seems to provide useful information about adherence using the reliable and valid SOCSI.

6.2 Introduction

There has been a considerable surge of adherence research in the last decade(497), yet some have cautioned against behaviour change studies that are not based on theoretical models.(411, 498, 499) Based on the researcher's intuition or implicit theory, these studies' results are often not reproducible, furthermore evaluating efficacy of interventions based e.g. exclusively on behaviour rather than theory, is problematic.(498) There is mounting evidence in other health domains that corroborate these claims: for example, two recent reviews indicated the effect of behaviour change was increased when interventions were based on theoretical concepts.(381, 500) Although adherence to sleep therapies is still in its infancy, there is emerging evidence to suggest the superiority of theory based studies and interventions in this field. A handful of studies in insomnia and sleep apnoea research have successfully examined the predictability of variables from social cognitive and stage based models.

6.2.1 The use of behaviour change theories in sleep research

A number of behaviour change theories have been examined in sleep research to date. Adherence to CPAP and CBT-I has been predicted by components of Wallston's social learning theory adapted for health,(304) and Bandura's social cognitive theory(231, 237, 286, 287, 296)

Two theoretical models that have been quite thoroughly explored within the CPAP and CBT-I adherence literature are the health belief model (HBM) and the transtheoretical model (TTM)/ stage of change model.

6.2.1.1 The Health Belief Model (HBM)

The HBM, developed in late 60s/early 70s(385, 434, 501), describes 6 distinct constructs as important in predicting implementation of new behaviour. The likelihood of the individual's behaviour is partly influenced by illness representations, such as the susceptibility to and severity of an illness and the evaluation of the perceived benefits of and barriers to change. Cues to action, which might be internal (e.g. the symptoms) or external (e.g. advice of health practitioners), might also lead to behaviour change. Two factors that have

recently been added to the model are health motivation and perceived control of the situation. This model has been utilised for predicting adherence to both CPAP(301, 308) and CBT-I(208)

Sage et al. 2001(308) and Olsen 2008(301) examined HBM variables adherences post and pre CPAP initiation respectively as outlined in chapter 1. Perceived benefits, when assessed before and after CPAP titration, were predictive of later CPAP use. Barriers and cues to action were only measured in Sage et al.'s study but only the former was an independent predictor of adherence. Self-efficacy and severity were not independent predictors in either study and susceptibility was in Olsen et al.'s, but not in Sage et al.'s study a predictor of later use.

Vincent, Lewycky and Finnegan applied this model to insomnia and CBT-I adherence.(208) They explored variables of the HBM and their predictability of adherence to stimulus control and sleep restriction (SC/SRT) components of CBT-I. In a sample of 53 individuals with insomnia undergoing a 6-week CBT-I program, the authors reported that fewer barriers to implementation of SC/SRT and less pre-treatment sleepiness were associated with increased subjective adherence based on sleep diary data. Again, lack of validated measures, resulted in not all constructs of the HBM being investigated. Furthermore, assessment of adherence to the treatment program was limited to self-report measures and sleep diary; both subject to bias by demand characteristics, if participants were aware that adherence was the main research focus.

6.2.1.2 The Transtheoretical Model (TTM)

With the call for increased attention towards psychological variables(369 see also chapter 2) and the shift in conceptualising adherence as a dynamic rather than dichotomous principle,(205, 495) the CPAP adherence field has welcomed the use of the transtheoretical model (TTM). Initially developed to understand behaviour change in smokers,(502-504), the application of this dynamic stage-based model is expanding to the contexts of other behaviour- particularly the cessation of unhealthy behaviour, such as substance use,(505) or unsafe sexual behaviours,(506) but also the implementation of healthy behaviour like exercise,(507, 508), mammography screening(509) or medication adherence.(510) With the intention of developing an integrative framework,

Prochaska and DiClemente identified concepts involved in behaviour change that were common to psychotherapies at the time. They suggested that individuals pass through five stages of change: pre-contemplation (no awareness of the need to change), contemplation (thinking about change), preparation (planning change), action (implementation of change) and finally maintenance (long-term implementation of change). These stages are associated with different profiles on the measures of decisional balance (pros vs. cons for change), processes of change (cognitive vs. behavioural strategies that are employed to move through the earlier and later stages of change respectively), and self-efficacy/temptation. In the early stages of change, self-efficacy and temptation are low and high respectively. Transition to higher stages witness as increase in self-efficacy and decrease of temptation. The transtheoretical model is described in greater detail in Appendix 8.

A number of studies have applied the transtheoretical model to the sleep field. One particular research group has scrutinized the utility of the TTM model in predicting CPAP use in both naïve patients in the short(286) and long term(237) and experienced CPAP users.(287) The first study in 2002(286) examined behaviour change variables prior to CPAP titration (baseline), 1 week and 1 month, and found that all but the baseline TTM variables were predictive of post treatment CPAP use. The same working group replicated these results with long-term CPAP use of 6 months.(237) Again, baseline measures explained little of the variance in subsequent adherence. In 2006, Stepnowsky and colleagues conducted a retrospective study examining TTM variables in experienced CPAP users (mean 2 years).(287) Despite measuring all concepts of the TTM (stage of change, decisional balance and processes of change), the authors seem to consider only the process of change and decisional balance index in their analyses, providing no information about stage of change and how this relates to CPAP adherence. This limitation makes it difficult to evaluate the use of the transtheoretical ***model*** in predicting CPAP use.

In the insomnia field, two studies to date have investigated the use of the TTM.(223, 511) One well-designed study explored the relationship between TTM variables and hypnotic discontinuation, providing insight into the usefulness of this model in understanding hypnotic tapering in individuals with insomnia.(511) As part of a larger randomised control trial (assessing hypnotic tapering with/without self-help non-pharmacological treatment for insomnia), the

authors evaluated the stages of change using an algorithm [see Appendix 8 for details on measurements], readiness to change with the University of Rhode Island Change Assessment,(512) decisional balance and self-efficacy in 53 participants over a 6-month study period. Baseline measures of all TTM variables (self-efficacy, readiness to change, and decisional balance) were not predictive of taper outcome group allocation. When comparing across groups, self-efficacy was the only measure that was significantly different between drug-free and non-drug free participants when measured mid-and post-treatment. When comparing those who were able to maintain their drug free status at 6-months and those who relapsed in the interim period between post-treatment and follow-up, self-efficacy and readiness to change were increased in the former group. At months 1, 3, 6, readiness to change was higher in those who did not relapse, as was self-efficacy at months 6. Of the 50 individuals analysed, 1 participant scored in the pre-contemplation, 17 in the contemplation, 32 in the preparation stage. Contrary to their hypothesis, there was no relationship between stage of change and group allocation. As the authors noted, this might be a result of both methodological and conceptual issues. The adapted measurements for stage allocation and readiness to change had not previously been validated in this population. Additionally, the transtheoretical model has been well described for 'problem' behaviours such as substance abuse, however hypnotics might not have the same negative connotation and the participant might have interpreted the term 'problem' to be the 'sleep problem'.

One other study has explored the use of the TTM in explaining adherence and attrition rates to an online CBT-I program.(223) Using an adapted version of a pain stage of change questionnaire, readiness to change was measured in 47 individuals with chronic insomnia to predict treatment attrition and adherence (defined as the percentage of participants using homework for at least 4 nights of the week). There was no significant correlation between readiness to change and attrition; the contemplation subscale was positively related to adherence to exercise recommendation and tapering of caffeinated beverages: both sleep hygiene components. Contrary to predictions, no other relationships were reported. The authors failed to mention an important conceptual issue with the readiness to change items used in their scale that might explain these negative results. It is important when considering readiness to change to acknowledge the

difference between motivation for change (or improvement) and motivation to participate in a particular treatment.(513) The following example from the contemplation subscale used by Herbert and colleagues, is more reflective of the former: *'I have recently figured out that it's up to me to deal better with my insomnia'*. Whereas a hypothetical item, such as *'I don't think making changes to my bedtime routine will lead to any improvements in my sleep'*, might highlight the individual's readiness (or lack of) to engage in the cognitive-behavioural intervention and reflect a stronger relationship with adherence. In addition, attrition rates and adherence were only related to a baseline measure of readiness to change. As explained above, measuring TTM variables prior to implementation of treatment, as seen in the CPAP trials, might be futile, if individuals have not been provided with ample opportunity to formulate beliefs and attitudes that might moderate their motivation. Lastly, of all transtheoretical model variables, this study was limited only to assessing readiness to change. Stage of change, decisional balance, process of change and self-efficacy are important constructs of the transtheoretical model that need to be assessed individually alongside important relationships between each constructs, to evaluate fully the transtheoretical model in predicting adherence to CBT-I.

In summary, there have been a number of studies that have perused theoretical health behaviour models to shed light on adherence to CPAP and CBT-I. However, several limitations can be identified with the above outlined studies:

1. Studies have fallen short of examining the overall impact of these theoretical models. Future studies should take care to include **all** components of the relevant theoretical model.

2. Insomnia or Sleep apnoea-specific scales that have been developed for this purpose have failed to include rigorous psychometric evaluation, especially patient-feedback, despite recent guidelines for patient reported outcomes to include assessment of patient understanding and readability of the questionnaire. (e.g. PROMISE(514)and FDA guidelines(515)

3. Adherence measures for CBT-I rely solely on self-reported questionnaires or sleep diaries (see also chapter 1). An objective measure of adherence to the behavioural components to CBT-I is required.

Although the health belief model and the transtheoretical model have most likely received equal amounts of attention in both health psychology in general and sleep research specifically, the use of the latter model might be more applicable for adherence research because of the natural transition to stage matched interventions for improving adherence.(500) Additionally, a more dynamic model, such as the TTM, might be more valid for understanding adherence to treatments for sleep disorders, when considering the characteristics of sleep and its disorders, such as insomnia.

Firstly, sleep is a dynamic process in itself. Some individuals with insomnia will present with extensive night-to-night variability in both the quantity and quality of sleep compared to good sleepers.(516, 517) Although this variability is targeted and effectively reduced during CBT-I,(233, 460, 518) any residual variability might influence participants' perceptions of treatment effectiveness. This might impact further on their motivation/readiness to adhere to treatment components. A model- such as the TTM- that accounts for these potential dynamic fluctuations might have greater potential in explaining variability in adherence.

A second important characteristic of the insomnia disorder is dysfunctional beliefs and attitudes about sleep (DBAS).(519) A strong endorsement of beliefs such as 'poor sleep is a result of a chemical imbalance' might be associated with reduced willingness to embark on non-pharmacological treatment options such as CBT-I. In this instance individuals might be less likely to recognise the problem as a result of poor sleep habits and less motivated to change these using strategies such as sleep restriction or stimulus control. This assumption is supported by a recent, thorough investigation into these cognitions. Carney and Edinger identified six of the 30 DBAS scale items that were amenable to change after treatment for primary insomnia, but had no 'good sleeper/insomnia patients' discriminative value. One possible reason for this paradoxical finding, as suggested by the authors, is that these beliefs are *'mediators of treatment change through increasing treatment adherence'*.(146, p.452) Again, a dynamic model, that accounts for some individuals not being

ready for change- potentially because of residual dysfunctional beliefs- might be useful in understanding adherence to non-pharmacological treatment options for insomnia.

Lastly, it is also important to highlight the subtleties of the insomnia specific behavioural/psychological interventions per se, when advocating the use of the transtheoretical model for our understanding of adherence. Behavioural components such as sleep restriction therapy and stimulus control require a patient to reduce initially their time in bed.(137, 520) This may seem counterintuitive to someone whose treatment goal is to increase the amount of total sleep time, especially when considering potential residual beliefs outlined above. Additionally, there is a general consensus that non-pharmacological treatments are 'safe' and are not associated with side effects like their pharmacological counterparts.(521) However, a recent study highlighted that individuals undergoing sleep restriction therapy may incur side effects, such as extreme sleepiness, fatigue and reduced motivation.(139) These debilitating experiences might negatively impact on individual's readiness to engage with treatment.

In summary, understanding adherence to non-pharmacological treatments for insomnia using the transtheoretical model seems logical after considering the characteristics of the disorder and its treatment.

6.2.2 Aims of this chapter

With the considerations outlined above, the goal was to develop an insomnia-specific SOC questionnaire that can be used to predict adherence to one single component of CBT-I: sleep restriction. After development of any instrument, cognitive pre-testing is a necessity in a sample of participants representative of the end-users. This will ensure that the questionnaire is comprehensible, that the intended cognitive processes are used in answering the questions, and that items generate a correct response (i.e. responses are correctly mapped onto the response categories presented). No previous author has published cognitive pretesting or any form of patient pre-testing for any existing stage of change questionnaire development. Patient involvement in psychometric validation instruments is essential(515, 522) to ensure the questionnaire measures what the developer intended. The next step was to establish further the psychometric reliability and validity of this scale in individuals undergoing treatment for their

insomnia. With limited information on optimal measurements of adherence, outlined in chapter 1, this study also investigated the use of novel techniques to assess adherence.

Thus, the following studies aim to

- Develop and cognitively pre-test a questionnaire to measure the components of the TTM (STUDY 1)
- Determine the concurrent and predictive validity the scale when applied to an insomnia population completing a sleep restriction program (STUDY 2)
- Test further subjective and objective measures of adherence to sleep restriction therapy adding to the currently limited information available (STUDY 2)

6.3 Study 1: Development and Preliminary Validation of the Stage of Change Scale for Insomnia(SOCSI)

6.3.1 Aim of study 1

The aim of study 1 was to develop and then specifically investigate patient-perceived validity of a stage of change scale specific for insomnia (SOCSI) .

6.3.2 Methods

6.3.2.1 Questionnaire Development

A team, which included two clinical psychologists specialised in sleep, and one research psychologist experienced in questionnaire development and psychometric evaluation, developed the questionnaire. Throughout this process three desired purposes of the questionnaire were discussed and agreed upon by the team during regular meetings. This scale should:

1. Assess all aspects of the transtheoretical model: stage of change, self-efficacy, pros/cons for change and processes of change.
2. Be generic, so it could potentially be applied to other fields of sleep (in addition to insomnia).
3. Be applicable to both research and clinical settings, thus it needed to be short and have good face validity.

The initial version of the SOCSI, was modelled on existing adherence stage of change questionnaires used in different clinical domains.(286, 523) Specific considerations for each component of the transtheoretical model were discussed:

6.3.2.1.1 Specifying the change of interest

In order to develop a scale that could be utilised as both a clinical as well as a research tool, open boxes were included for individuals to enter the specific behaviour to be changed (e.g. changes in get up and bed time). This enabled the scale's generalisability to other domains of interest, both within insomnia as well as other health areas.

6.3.2.1.2 Stage of change

As outlined above, a variety of formats have been used to assess stage of change, these vary from continuous scale measures, to single item (forced choice) format and algorithms. There was consensus that the latter options (single item/forced choice and algorithm) would be most appropriate, because of their simplicity, in accomplishing the aims outline above. Thus two versions of the scale were developed to accommodate both the single item and algorithm option. These were then subjected to further scrutiny by the participants in this study (see below). Figure 10 shows the difference between these two options (note, this is taken from the final versions post cognitive pre-testing).

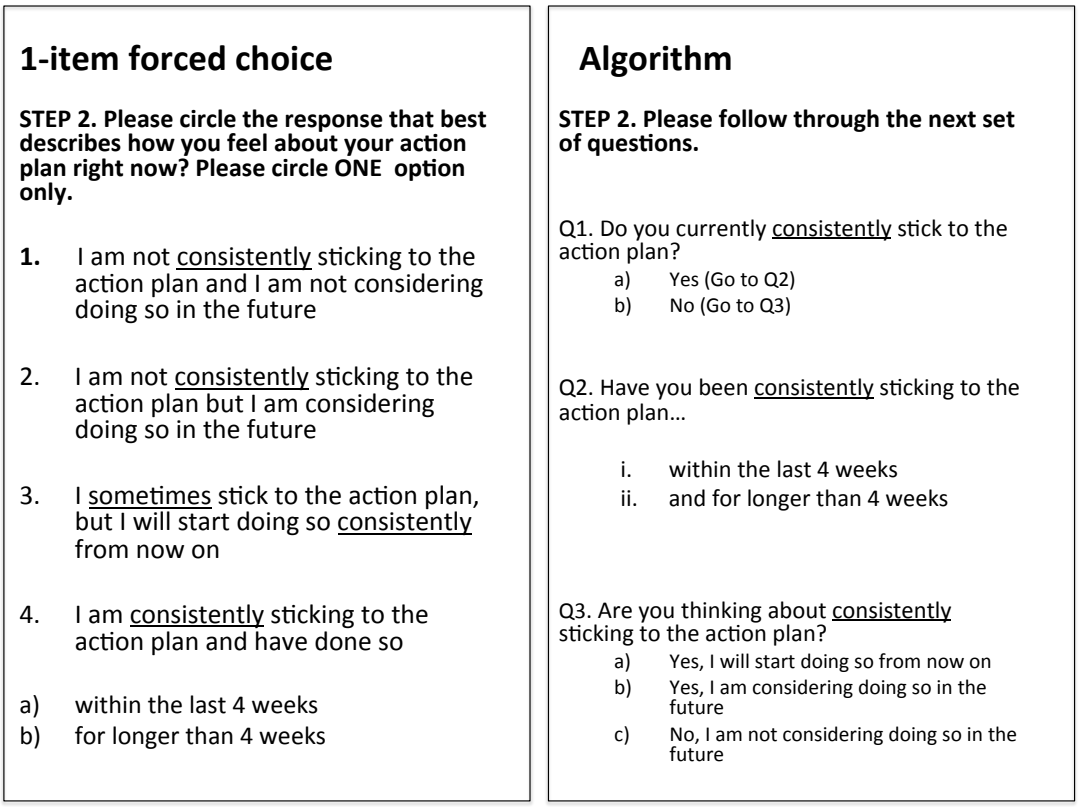


Figure 10: Forced Choice vs. Algorithm

One consideration discussed, was the use of a baseline measure, which assessed stage of change before treatment had been initiated. Previous stage of change scales e.g.(286) have included the action and maintenance stage at baseline assessment. This seems inherently redundant, if an individual, who has not yet had a titration study or even seen the machine , is asked to make a forced choice between *'I currently do not use CPAP, but I am thinking about starting to use CPAP in the next 2 weeks'* [contemplation] AND *'I currently use CPAP nightly and I have begun doing so within the last 2 weeks' [action].* The

baseline SOCSI (to be administered before treatment initiation) therefore only included three stages: pre-contemplation, contemplation and preparation. All other items remained identical.

Lastly, decisions were made in terms of what constituted each stage in this population. These decisions were based on existing descriptions of the stages as well as clinical experience with the insomnia population. The pre-contemplation stage described an individual who was not willing to consistently adhere to components of CBT-I. Contemplation and Preparation were described as individuals who were not consistently adhering to the treatment, but thinking and planning on doing so respectively. Some scales separate these stages by indication of some behavioural changes in the preparation change (e.g., 286) however because there is little information about what optimal/partial constitutes adherence to CBT-I, it was decided to remain with a more intentional/temporal distinction (considering doing so in the future vs. will start doing so from now on). Lastly, action and maintenance stages were separated by the temporal distinction of 4-weeks, despite the original description of 6 months as a differentiating time between the action and maintenance stages. This time frame reflects, more appropriately than 6 months, the relatively immediate improvements in sleep reported in most clinical trials of CBT-I. Additionally, some have described the implementation of a booster sessions after CBT-I within these short time periods, to prevent relapse(e.g., 233, 524)

6.3.2.1.3 Self-efficacy

Because one of the aims was to develop a short questionnaire with good face validity, easily administered within the clinical setting, a single 5-point item for self-efficacy was used. Although this concept is considered multi-dimensional,(340) a 1-item scale has been shown to be a reliable predictor of adherence to CBT-I.(231)

6.3.2.1.4 Pros/Cons for Change and Processes of Change

As there are currently no validated measures of decisional balance and process of change for adherence to CBT-I, the questionnaire was designed to generate patient's accounts of the pros and cons, and processes of change, using the open box response format. The secondary gain of this patient-generated information

is the further development of psychometrically more *reliable* measures to assess the decisional balance and processes of change.

6.3.2.2 Participants

Participants were recruited if they were currently, or had previously completed a CBT-I program at the Woolcock Institute for Medical Research, Sydney and University of Glasgow Sleep Centre, UK. Participants included a mixture of both clinic patients and research participants. Only individuals who were between 18-65 years and proficient in the English language were approached for participation in this study. Ethical approval was obtained from the local ethical committees NHS Greater Glasgow and Clyde, and Sydney Local Health District Ethics Review (RPAH Zone).

6.3.2.3 Cognitive Pre-testing

Cognitive pre-testing is a methodology, which can assess patient acceptability and comprehension of survey questionnaires as part of face validity testing.(525) Two main methodologies can be used in isolation or combination, think-aloud and probing.

6.3.2.3.1 Think-aloud process:

This methodology can be administered in concurrent and retrospective fashion. Within this approach the individual thinks-aloud during or after filling out the questionnaire. Rather than just responding to an item, the individual is asked to verbalise all thoughts involved in the process of responding to a particular questionnaire item. The interviewer takes note and records the responses with no or little probing.

6.3.2.3.2 Probing:

With this methodology, the interviewer uses certain probes to follow the responders thought processes from reading and processing a question (or instruction) through to formulating a response. Probes can be proactive or reactive. Proactive probes are generated by the interviewer and can be anticipated, hence prepared beforehand, whereas spontaneous probes are devised in reaction to novel and important responses or themes emerging during the interview. Both proactive and reactive probes can be standardised i.e.

elicited in response to certain behaviour of the interviewee (e.g. if subject pauses before answering, the interviewer might ask about the reason for the pause) or unstandardised, i.e. emergent (spontaneous responses to the interviewee's unique behaviour). The current study involved a mixture of both procedures to cognitively pre-test this insomnia specific SOC questionnaire using the following procedure:

6.3.2.4 Interview Procedure:

After obtaining consent (consent forms and patient information sheets are similar in format and content to those in Appendices 2, 3 and 6), general procedures of cognitive pretesting were explained. The participant was then asked to fill in each questionnaire (all 3 versions: baseline, forced-choice, algorithm) without input from the interviewer and encouraged to mark any comments on the questionnaire. To minimise order effects, the presentation of the forced-choice and algorithm order was randomised. The interviewer then progressed through through each questionnaire step by step (see Appendix 9 for an excerpt of the interview protocol), encouraging the individual to think aloud as much as possible. Conditioned probes (Appendix 10) were prepared prior to the interview. After the semi-structured interview, the individual was encouraged to make any general comments regarding the questionnaires. All interviews were audio-recorded.

6.3.2.5 Data analysis

A detailed review of the audio-recorded responses to each instruction, question and each response option was conducted at the end of each interview. The semi-structured interview schedule and questionnaire were changed in an iterative process as novel themes emerged that required further exploration with subsequent participants or as changes were made to the questionnaire (i.e., rewording or reformulation due to comprehension problems, alternative wording suggestions by patients, or restructuring of the instrument).(525) No individual change was made that was not subsequently cognitively tested by at least 2 participants. The SOCSI was amended iteratively by two researchers of the development team, with the theoretical underpinnings of the model in mind, and interviews continued until no new issues emerged (saturation).

6.3.3 Results

Saturation was reached after 13 individuals (mean age=39, mean education level= 16, range12-19) were interviewed. Minor changes to the questionnaire were confined to issues with layout, confusing wording, and the use of jargon.

The more substantial changes made to the questionnaire were 1) the term used to describe the behavior change of interest and 2) the time frame and distinction between stages of change items 3) terms to describe pros/cons and processes and 4) the term to describe 'adherence', see Table 13 for further descriptions.

Table 13: Major Changes to SOCSI after Cognitive Pretesting

Changes	Changed from	Changed to	Rational for Change
1. Term used to describe the behavior change of interest	**are you sticking to 'the above'** (in reference to the patient's response in the open box format)	**are you sticking to the action plan**	The use of the word action plan was suggested by one participant, and preferred by subsequent participants over vague alternative options (e.g. 'the above' or 'sleep strategy'). This term was better understood and generally reflected better the notion of actively doing something.
2. Distinction between the preparation and contemplation stage	'**Planning to consistently stick to the above in the near future** '(preparation) vs. '**considering consistently sticking to the above'** (contemplation)	**'I will start doing so from now on'** (preparation) vs. '**I am considering doing so in the future'** (contemplation)	The poor distinction between the two stages is often quoted as a possible reason limited difference in behavior change between these two stages. This was also reflected in the cognitive interviews, and individuals struggled to identify differences between these two items. One participant suggested the addition of 'from now on' and the use of the word 'will' rather than 'plan to' would reflect a more stark contrast to the contemplation stage.
3. Terms used for			
Pros/cons	**Advantages/Disadvantages**	**Advantages/Disadvantages**	Some participants suggested changing this to pros/cons, however the majority of individuals preferred the advantage/disadvantage distinction.
Processes	**'what strategies do you have to help you…'**	**'what can you do to help you stick to the action plan'**	This change was more comprehensible to individuals, and also reflected a more dynamic nature underlying these processes, which is in line with the TTM model.
4. Terms used to describe adherence	Sticking to	Sticking to	Although this term remained unchanged, it was important to determine the most acceptable term for adherence. All participants favoured this term over alternative suggestions (adherence, compliance, follow the treatment regime etc.)

One individual felt the need for a response reflecting a stage between the preparation and contemplation stage. However this was inconsistent with the transtheoretical model, and thus this was not considered in this adaptation phase of the questionnaire.

Overall, interview responses indicated good face and content validity, and no other missing responses were indicated by individuals. Interestingly, three participants remarked that completing the questionnaire made them more aware of their motivation to use sleep strategies.

'I didn't answer straight away, I had a little think about generally how confident I was…and thought about how I had been performing thus far, which is not very good, so that has made me less confident'

'it's good, it's a challenging question [on the decisional balance], *it's always good to think about why you are doing these things that you are doing'*

'Its a great idea to have this sort of questionnaire, because it does get you to look into the deep dark tea time of the soul and think, well hang on it is all up to me'

These quotes provide preliminary evidence for the accomplishment of one of the aims: to develop a tool that could be used in clinical setting to monitor and potentially improve stage of change.

At the end of each cognitive interview, the participants were asked which version they preferred. The algorithm version was the preferred questionnaire format (n=8/13) and this was used to further validate the SOCSI.

6.3.4 Preliminary Conclusions

This cognitive pretesting study presents important patient-reported validity data. Results suggest that the SOCSI is valid and comprehensible to individuals. The brevity and easy of administering the SOCSI suggests it's potential for being an integral of the therapeutic process in daily clinical practice. The final questionnaire is presented in Appendix 11 and further validated in Study 2.

6.4 Study 2: The Stage of Change Scale for Insomnia (SOCSI)- A new scale to monitor stage of change during sleep restriction therapy for insomnia

6.4.1 Aim of study 2

Based on the results of the previous study, the final questionnaire was used with a group of individuals with insomnia undergoing a sleep restriction program. The primary aim of this study was to assess further the reliability and validity of the SOCSI: is the questionnaire measuring what it is set out to measure and can it reliably do so. In order to examine the scale in relation to these criteria the following study was conducted.

6.4.2 Methods

6.4.2.1 Participants

Participants for this study were recruited at the University of Glasgow Sleep Centre using routine recruitment methods: posters were distributed in public areas (such as libraries, GP surgeries, sport and community centres). Additionally individuals who were on the sleep centre's research database and who had consented to being contacted for research purposes were approached about participation in the study. Suitable individuals- as determined by preliminary screening interview used at the UGSC (see Appendix 12)- were followed up by the principal study investigator for a thorough screening interview as outlined elsewhere.(465) Inclusion was evaluated against established research diagnostic criteria for primary insomnia, see appendix 1.(RDC 92) Individuals had to present with a difficulty falling asleep, staying asleep or waking too early, despite adequate sleep opportunity. The sleep problem, lasting for a minimum duration of 1 month, was also associated with difficulties in one of the core daytime functioning areas outlined in the RDC criteria. Further requirements for inclusion were:

- Between 18-65 years
- Naïve to behavioural treatments for insomnia and currently not

receiving any pharmacological treatment for their insomnia (those who had successfully tapered off their hypnotic medication for longer than 2 weeks [under guidance by their general practitioner] were eligible for the study.)

- Fluent in English and able to understand, participate and comply with the requirements of the study procedures.

Participants who had a co morbid psychiatric or medical disorder, which was temporally related and/or attributed to their sleep problem(92), were excluded. Additional exclusion criteria were suspected co-morbid sleep disorder (circadian rhythm, sleep apnoea, narcolepsy, restless legs syndrome) and an irregular sleep/wake pattern that would interfere with the treatment program (e.g. nightshift).

6.4.2.2 Study Procedure:

This study was approved by the NHS Greater Glasgow and Clyde ethics committee (consent forms and patient information sheets are similar in format and content to those in Appendices 2, 3 and 6). After obtaining participant consent, individuals were asked to complete a sleep diary for 7 days prior to arrival at the sleep centre, where the sleep restriction treatment was then delivered to groups of 2-3 people. The sleep restriction treatment protocol (see Appendix 13) was adapted from the original treatment description(137) and the University of Glasgow CBT-I protocol, described elsewhere.(207, 464, 465). Sleep Restriction (or perhaps more accurately bed restriction and from hereon referred to as such), involves curtailing the time spent in bed to the individual's average sleep time obtained from the diary. A morning rising time is set according to personal and circadian preference. The average sleep time is subtracted from this rising time to establish the threshold time, when or after which individuals were allowed to retire to bed. Individuals are instructed to only be in bed between these allocated times. Subsequent weekly titrations are made to the window according to the following criteria: 15 minutes increase to the bed window if weekly sleep efficiency ratings is more than 90% [sleep efficiency being the percent of time spent in bed asleep]; no change if sleep efficiency was 85-90% and a downward titration by 15 minutes if sleep efficiency was below 85%. The minimal bed window was 5 hrs.

The acute treatment phase in this study was 4 weeks. In the first session, participants were introduced to the bed restriction program, along with a brief introduction to sleep/wake processes and treatment rationale. Individual bed windows were calculated based on the 7-day baseline sleep diary and subsequent titrations were made each week. The first 3 sessions included discussion of the sleep restriction rationale, calculating sleep efficiency and titrating the bed window, potential trouble shooting and questionnaire completion. The content of the last two sessions and 3-month follow up session was reduced to bed window titrations and questionnaire completion. Figure 11 and Figure 12 for an outline the study and treatment sessions respectively.

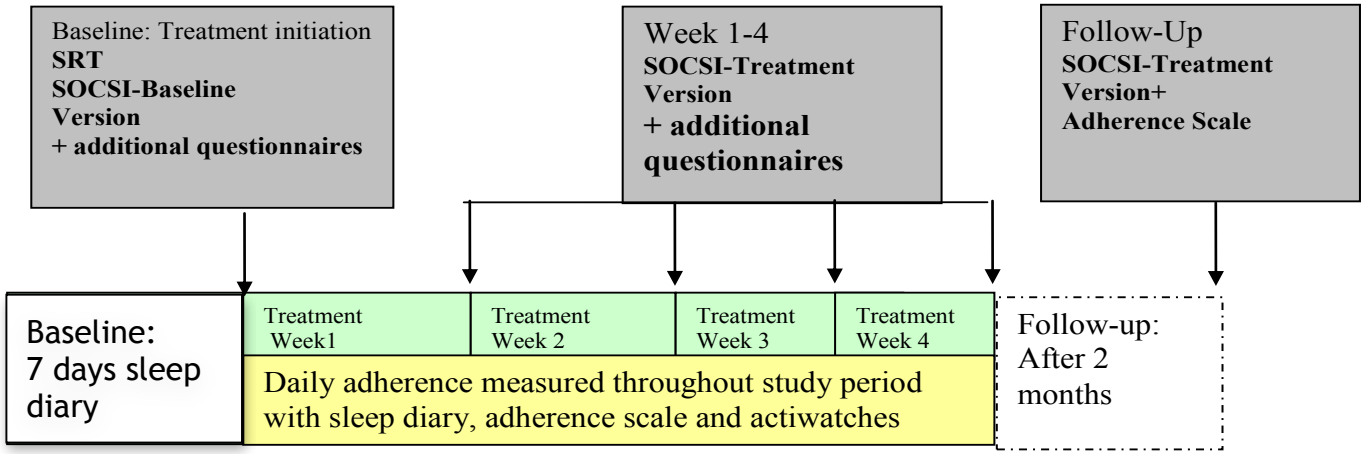


Figure 11: Study Outline

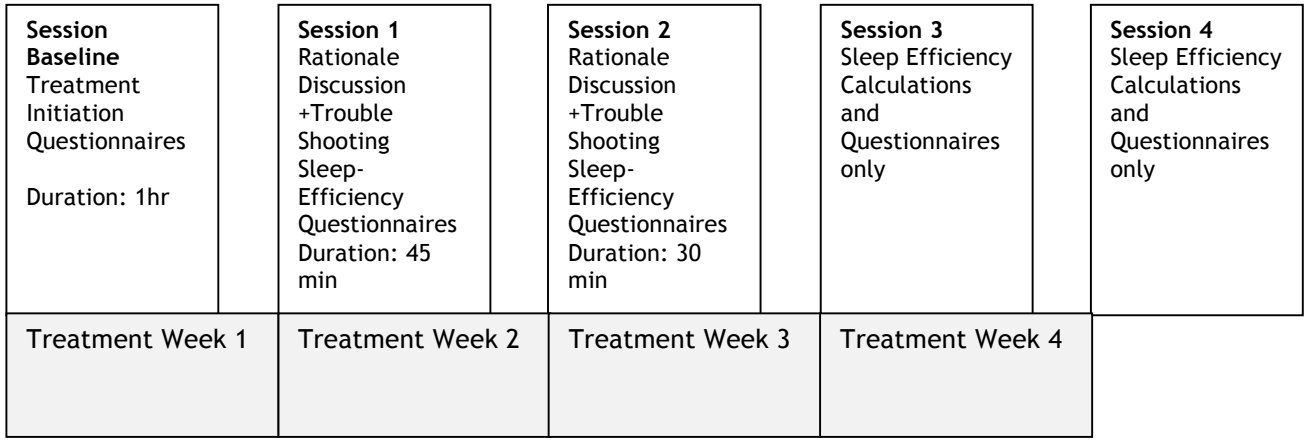


Figure 12: Outline of treatment sessions

6.4.2.3 Measures:

6.4.2.3.1 Outcome measures of sleep and mood

The following measures were administered to assess sleep and mood outcomes:

6.4.2.3.1.1 Sleep Diaries:

The Sleep Diary for insomnia(465) is a daily assessment of the quality, quantity, and timing of sleep episodes. The sleep diary in this study was slightly adapted from the original to include information on napping, overall sleep quality and early morning awakenings (EMA) in addition to the usually reported subjective sleep outcome variables: Wake-after-Sleep-Onset (WASO), Sleep Onset Latency (SOL), Total Sleep Time (TST), Sleep Efficiency (SE), Time in Bed (TIB) and Sleep Quality (SQ), see Appendix 14.

6.4.2.3.1.2 Insomnia Severity Index (ISI)

The ISI(102) is a 5-item questionnaire used to assess the severity of the insomnia symptoms (onset, maintenance and early morning awakenings); daytime and quality of life impairment attributed to the sleep problem, as well as dissatisfaction with sleep and distress associated with insomnia. Used as both a clinical assessment tool and research outcome measure, the total score can be used to classify individuals into varying degrees of severity: No clinically significant insomnia (0–7); Subthreshold insomnia= 8–14; Clinical insomnia (moderate severity)= 15–21; Clinical insomnia (severe)= 22–28. The reliability (Cronbach's coefficient alpha=.74) and validity (moderate correlations with sleep diary measures and established sensitivity to change with cognitive behaviour therapy) have been established,(526) justifying its widespread use.

6.4.2.3.1.3 Pittsburgh Sleep Quality Index (PSQI)

The PSQI(105)] is a measure of sleep quality and sleep disturbances over a 1-month period. It has been used as an outcome measurement in insomnia studies as well as a valid tool for distinguishing good and poor sleepers. The total score (0-21) is made up of 7 subscales (Sleep duration, sleep disturbance, sleep latency, daytime dysfunction due to sleepiness, sleep efficiency, overall sleep quality and medication use). Higher scores indicate poorer sleep quality. In a

validation study Backhause et al. used the cut-off for poor sleep quality (total score>5) on the PSQI to correctly identify individuals primary insomnia with a sensitivity of 98.7% and good sleepers with a specificity of 84.4%. In the primary insomnia group, test-retest reliability was good, as was the internal consistency of the scale, with a Cronbach's alpha of 0.85. Items of the PSQI correlated with the respective sleep diary entries.(527)

6.4.2.3.1.4 Hospital Anxiety and Depression Scale (HADS)

Originally developed by Zigmond and Snaith, the HADS was developed as a tool for identifying and quantifying depression and anxiety symptoms in hospital patients.(528) It has now become a well-used scale extending beyond the initial patient group. The questionnaire consists of 14 items from which the 7 anxiety items and 7 depression items make up the respective subscales. A review of validation studies indicated a mean Cronbach's alpha of 0.83 for the anxiety subscale and mean internal consistency ratings of 0.82 for the depression scale. Across studies the scale showed good sensitivity and specificity (around 0.8) and strongly correlated with other validated depression and anxiety scales.(529)

6.4.2.3.1.5 Dysfunctional Beliefs and Attitudes about Sleep Short Form (DBAS-16)

The DBAS-16(530) is a measure of sleep-related dysfunctional thinking, adapted from the original 30-item scale.(519) Individuals are asked to indicate the level of agreement on sixteen statements; a strong endorsement of these statements is suggestive of dysfunctional beliefs and attitudes about sleep. This 16-item version is comprised of 4 subscales that reflect individual's misconceptions about 1) perceived consequences of insomnia 2) worry/helplessness about insomnia 3) sleep expectations and 4) medication. The scale has acceptable internal consistency (Cronbach's alpha between 0.77 and 0.79) and adequate test-retest reliability across a 2-week interval (r= .83)(530).

6.4.2.3.2 Stage of Change Questionnaire

The SOCSI was administered at each observation point (baseline, week 1- 4 and follow-up session). The final version obtained through the cognitive pretesting study outlined above was used.

6.4.2.3.3 Motivation Scale (MOT)

The motivation scale is a twelve-item questionnaire that represents dimensions occurring in the natural language to describe motivation to change.(531) The three-factor structure reflects dimensions of patient motivation to change: importance, ability, and commitment. Internal consistency of the total scale was good (Cronbach's alpha=.86). The authors noted redundancy amongst the 12 items and identified three items that best reflected each factor, however the internal consistency of the 3-item version was low, ranging from 0.39 to 0.69 across different sites. The 12-item total scale was sensitive to change after motivational interviewing style treatment for substance misuse. One limitation of the scale noted by the original authors was the ceiling effect, with 43% of the sample choosing the highest score 10 on all 12 items.

6.4.2.3.4 Potential Predictors of Adherence

Additional baseline measures were assessed to examine their relationship with adherence. This included a measure of sleepiness as assessed by the Epworth Sleepiness Scale [ESS(5)] a measure of fatigue using the Flinders Fatigue Scale [FSS(532)] and the illness perception questionnaire-revised [IPQ-R(533)] The ESS is a measure of the likelihood of an individual falling asleep in 8 situations with a total score ranging from 0-24; a score of ≥ 11 is indicative of excessive daytime sleepiness. Test-retest reliability of this scale is good across a 5-month period (r=0.85) and internal consistency is excellent (Cronbach's alpha=0.88).(534). Consisting only of 7 items the FFS is a brief measure to document the individuals daytime fatigue in different situations. Scores range from 0-32 with higher scores indicating greater fatigue experienced. The original paper reported excellent internal consistency (Cronbach's alpha=.91) and non-significant correlations between the ESS and FSS support the differentiation between the constructs fatigue and sleepiness. The IPQ-R measures how individuals make sense of their condition and the treatment on the basis of 7 dimensions Timeline acute/chronic, Timeline cyclical, Consequences, Personal control, Treatment control, Illness Coherence, Emotional representations (expanded from Leventhal's original five /illness perceptions.(473) The original authors reported good internal consistencies for each subscales ranged from (0.7-0.89) and adequate test-retest reliability across a 3-week (r=.5-.7) and 6 month period (r=.5-.8).

6.4.2.3.5 Adherence Measures

As outlined in the introduction, several subjective methods have been developed to measure adherence to components of CBT-I. These vary from spousal/therapist rated or self-report questionnaires to adherence scores obtained from the sleep diaries. Only one study to date has described the objective measurement of adherence using actigraphy.(228) The present study reports on the use of 3 types of adherence measurement: self-report, diary and actigraphy.

6.4.2.3.5.1 Self-report adherence questionnaire

Well-validated self-report questionnaires to assess CBT-I adherence are sparse. One study(208) adapted the five-item Medical Outcomes Study Specific Adherence Scale(212) to assess adherence to sleep restriction and stimulus control. Three of the five items assessed individuals' adherence to the treatment plan (e.g. I was unable to do what was necessary...") however two of the items assessed 'ease' and 'difficulty' in adhering to the doctors recommendations, aspects that are not truly reflective of adherence: one could find something hard, but still do it. This might explain the moderate Cronbach's alpha of .78 reported by the original authors. For the purpose of the present study, a self-report (bed restriction specific) adherence questionnaire was developed assessing adherence at a general level, as well as adherence to threshold and rising time on both weekends and weekdays. Thus 5 measures were obtained from this questionnaire: total adherence (sum of all items); and average adherence scores for the two threshold, two rise, two weekend and two weekday items. See Appendix 15 for the questionnaire.

6.4.2.3.5.2 Sleep Diary

A number of studies have made use of the sleep diary data, with some using the absolute values to create scores of minute deviation and/or consistency in rising time/threshold time(208, 213, 214); and other creating composite scores that depict a more conservative estimation of non-adherence.(231) A combination of these measurements was used in this study to depict the most appropriate assessment of non-adherence:

1. Composite Score of minute deviation

2. Rising Time Deviation Variance

3. Threshold Time Deviation Variance

See Table 14 for a detailed description of each measurement.

Table 14: Description of Adherence Measurements

Composite Score (CS) for threshold and rising times	**Previous measurements:** The composite score was adapted from existing measures reported in the literature. Out of Riedel and Lichstein's strategies for assessing adherence to bed restriction, one measure described absolute deviation in minutes from the agreed time in bed. Bouchard and colleagues computed a composite score based on 7 adherence behaviours. Two of these adherent behaviours concerned the bed restriction component of CBT-I. Points from the total composite score were subtracted, if the individual went to bed more than 15 minutes before the threshold time and got up more than 30 minutes after the rising time. **Problem with these techniques:** The absolute deviation in Riedel and Lichstein was not related to outcome. The composite score reported in Bouchard included assessments of components that might have not needed implementation with improvements in sleep (e.g. removing oneself from bed when awake for more than 30 minutes) and this could potentially have biased the score. These limitations were considered when deciding on how to calculate optimally the composite score in this study. **Calculation of the composite score:** The CS was computed from the absolute deviation from the rising and threshold time. Absolute deviation was considered as any time before the threshold and any time after the rising time. Bed and get-up times that fell within the agreed window were scored as zero, so the absolute value represented true non-adherence. The score for each rising and threshold deviation time ranged from 1-3, a minute variation of 1-15 indicated an CS of 1, between 16-30 was given a score of 2 and anything above 30 minutes was given an CS of 3. Thus for each week an individual could obtain a score between 14-42, with higher scores indicating poorer adherence (larger minute deviation). Weekly and total 4-week treatment scores were obtained.
Rising Time Deviation Variance Threshold Time Deviation Variance	**Previous measurements:** Riedel and Lichstein not only computed the absolute deviation, but also the consistency of time in bed/rising time. A moderate association between this measure and outcome indicated that consistency in the bed times was also important, if not more than absolute deviation from the bed restriction window. **Problem with these techniques:** Firstly, the association between consistency and outcome has not been replicated by others(214). Secondly, this should not be considered a true measure of adherence, as individuals undergoing bed restriction are instructed to go do bed on **or after** the threshold time. A true measure of adherence would need to take this into account. **Calculation of the deviation variance score:** Instead of calculating a measure of time in bed consistency, it was decided to compute a measure of variance in non-adherence. So, rather than examining whether participants were consistent in their bed/get up times, it was examined whether they were consistent in their non-adherence. From the absolute deviation (only times before threshold and after agreed rising time) standard deviations were created. For example, someone who got up 30, 60 and 120 minutes after their rising time, had a rising time deviation variance of 46 minutes. Higher values indicated greater variance and thus poorer adherence. Weekly and total 4-week treatment scores were obtained.

6.4.2.3.5.3 Actigraphy

Actigraphy wristwatches (Actiwatch, Cambridge Neurotechnology Ltd., Cambridge, UK) worn on the non-dominant wrist were used to measure participant's "objective" adherence to their threshold and get up time. Individuals were asked to wear the actiwatch for the duration of the 4-week intervention and were asked to depress the event-marker button on the face of the watch to indicate each bed and get-up time. Actigraphy watches are equipped with a piezoelectric accelerometer and memory card to enable the recording and storing of physical movement data. Thus, compared to electroencephalogram (EEG), actigraphy does not measure sleep per se, but inferences are drawn from the lack of movement about the individuals' sleep/wake pattern. Nevertheless, a large evidence-base is accumulating to provide validity of actigraphy when compared to polysomnographic recording.(535) Information about the intensity and duration of the activity is stored epoch by epoch. Rather than sleep/wake patterns, the outcome measure of interest in this study was time in bed, which enabled the measurement of the extent to which individuals adhered to their agreed bed window. A time stamp for threshold (time to bed) time and rising time was established by electronic analysis of movement. Actiwatch data were imported into Philips Respironics Inc Co. software and time stamps were identified by the software's default settings for establishing rest periods (time before sleep and wake onset, demarking time in bed). Additionally, the depression of the event marker on the face of the watch provided information about individuals' bed and get-up times. The actiwatches were set to an epoch length of 1 minute, to obtain information over the 4-week intervention period. This technique has previously been reported in the use of objective adherence to a sleep restriction therapy in good sleepers.(228) The computation of the composite scores and rising and threshold time deviation variance for the marker and movement information was identical to that of the sleep diary.

Sleep Diary and Actiwatch adherence measures were only available for the 4 weeks treatment phase, as these measures relied on knowledge of weekly-agreed bed window, which was not available at follow-up.

6.4.2.4 Data Preparation: Missing and Excluded data for sleep diaries and actigraphy

In both the sleep diary and actigraphy methods, data were excluded when less than half of the data points were present. When this criterion was used for the weekly calculations (i.e. <4 of the 7 entries), 9 weeks of the total 972 weeks (4 treatment weeks per 27 people * 3 adherence measurements * 3 adherence methods), equating to 0.009 % of excluded weekly entries. Four-week averages created over the 28 days, only one individuals actigraphy threshold variance data were excluded (i.e. less than 14 entries). This data were excluded casewise in subsequent analysis. According to the Philips Respironics report,(536) the automatically determined rest periods require corroboration by visual examination of the data. An arbitrarily defined cut off of 2hrs was used to determine outliers (when the software determined rest period was more than 2hrs before or after the agreed threshold and rising time respectively). In this case the marker entry was imputed. This occurred for 84 entries (0.08%). If no marker existed the entry was deleted. (40 get up times and 52 bed times= 0.04 and 0.05% respectively) There were no weekly data points excluded for the movement data.

6.4.2.5 Statistics Analyses

The core focus of this study is the psychometric evaluation of the SOCSI and its association with adherence. Because this is explored in the context of a sleep restriction trial, preliminary "assumptions" needed to be met. Firstly the treatment intervention was expected to confer improvements in sleep and mood related outcome measures. Secondly, with limited consensus on optimal measures of adherence, the individual adherence variables and their relationship to outcome were examined. The four outcome measures assumed to be related to increased adherence were defined a priori as sleep diary obtained sleep efficiency (SE), wake after sleep onset (WASO), sleep onset latency (SOL) and insomnia severity index (ISI). This enabled further evaluation of the adherence variables that most strongly related to outcome. The results section is separated into three components: 1) treatment oucome and adherence measures (preliminary "assumptions") 2) psychometric evaluation of the SOCSI and 3) content analysis of the open-box response formats. The subsequent paragraphs outline the analytic approach for each of the three components. Continuous variables are presented as mean (SD) and p values of <0.05 are considered statistically significant. Non-significant trends (p<0.1) are reported; statistical results with p-values > 0.1 are not further specified unless deemed theoretically interesting, in which case they are further outlined in the appendix.

6.4.2.5.1 Treatment outcome and Adherence Measures

Repeated measures ANOVA was computed to examine treatment related effects across post treatment and follow-up; Bonferroni correct post-hoc comparisons were conducted to follow-up significant main effects. The same statistical test was adopted to examine differences between the adherence methods (sleep diary, actigraphy marker, actigraphy movement) for the composite score. Although some of these variables were non-normally distributed, the ANOVA has been shown to be robust measure of non-parametric data in sample sizes of more than 25.(537) Normality was assessed with the Shapiro-Wilk test (p>.05) and visual inspection of histograms.(538) Pearson's correlations (Spearman's where data were not normally distributed) were used to examine the relationships between adherence*outcome and baseline variables [sleepiness, fatigue and illness representations] *adherence. Analysis related to the

reliability and validity of the SOCSI is outlined below.

6.4.2.5.2 Psychometric Evaluation of the SOCSI:

6.4.2.5.2.1 Validity

Criterion Related Validity was established by examining the relationship between stage of change and concurrent adherence and motivation assuming that those in higher stages would present with better adherence and increased motivation (concurrent validity). The relationship between stage of change at baseline and adherence over the 4 weeks treatment phase will provide evidence to support the predictive validity of the SOCSI. Because of the small sample size and limited spread of frequency counts across the stages, the contemplation and preparation stage were merged. Adherence and motivation scores were contrasted between the action and pre-action stages using independent samples t-test, or Mann-Whitney test where data were non-normally distributed.

6.4.2.5.2.2 Test-retest reliability

Although usually a 2-8 week window between measurements is recommended for test-retest reliability statistics(539), this is inherently problematic when examining stage of change as the model implies dynamic changes across time-points and some studies have even indicated change as short as three days.(540) It is thus usually uncommon for scales measuring stage of change to assess test-retest reliability. Although some have attempted this over a 2-week period(541, 542), this methodology is inconsistent with the model. Changes across this time period could reflect true stage transitions, rather than an unreliable measure. Others have used a much shorter time frame, for example Donovan et al(543) assessed stage of change in one session, with other questionnaires intervened between the two time points. This methodology might also be unsound, because responses to these questionnaires could lead to changes in individuals' readiness to change. Therefore, this study used a shorter time period akin to Donovan et al.'s approach, however intervened with a short 10-minute arithmetic task, to prevent individuals from rehearsal/rumination of their responses to the SOCSI. Test-retest was measured at week 1 only. Details of the arithmetic distractor task can be found in Appendix 16. Intraclass Correlation Coefficient between time point 1 and timepoint 2 for both the stage of change and self-efficacy item

was calculated to determine test-retest reliability.

6.4.2.5.2.3 Content Analysis of the SOCSI's open-response questions

Three questions in the SOCSI were open-response format, generating participant's perceptions on:

- The advantages of sticking to the action plan
- The disadvantages of sticking to the action plan
- Things that help sticking to the action plan.

These questions overlapped with the decisional balance (pros and cons for change) and processes of change as outlined in the transtheoretical model. Participant generated responses were analysed using content analysis(544). Each thematic unit, that is each object that is considered conceptually and logically different from other objects, was coded and grouped into semantically meaningful categories. Generally, only one-unit responses were generated for each question, however a number of participant responses produced multiple separate units within the response. Because the question was phrased to generate multiple responses e.g. 'what are the advantage**s**', each unit was identified and counted as an individual unit with equal weightings to one-unit responses. Relative frequency counts ((category/all categories generated)*100) for each thematic category were subsequently produced for each observation point (baseline, week 1-4 and FU) as well as for all time points collectively. The analysis of the data generated for the process of change were guided by the theoretical underpinnings of the transtheoretical model (10 cognitive and behavioural processes of change outlined in Appendix 8). A second reviewer, independent to the study reviewed the identification of the themes.

6.4.3 Results

6.4.3.1 Patient Characteristics

Thirty individuals were recruited into the study. At 4 weeks post treatment, there were 28 completers and 2 drop outs. Reasons for drop outs were overwhelming side effects (n=1) and participation in the study was untimely (n=1). Of the 28, one person was exlcuded because of nocturnal actigraphy movement output that was visiually indistinguishable from the diurnal output. This individual was referred to a neurologist for further investigation. At the 3

month follow-up stage, a further 2 participants were lost to follow-up (1= uncontacatable; 1=overwhelming negative side effects of treatment). Their data was imputed by the last observation carried forward (LOCF). Although alternative methods such as using mixed-effects models are prefered over this option(545), the present study was not a randomised control trrial, and the LOCF method for only 2 cases was not determined as biasing the results. Table 15 presents the patient demographics of the 27 participants who completed the treatment phase. The sample's mean age was 48 years, ranging from 19-65 years. The 22 females and 5 males were well-educated, with an average education of 15 years. The majority of the sample were either single (n=8) or married (n=8), whilst others had a live-in-partners, were divorced, separated, withowed or whose partner lived seperately (n=11). Fourty percent of the sample lived alone, whilst 55% lived with a spouce, children, other family number or a room mate. Whilst the majority of the sample indicated they were either white-british or white-scottish (70%), one individual selected asian-pakistani and seven individuals did not provide information about their ethnicity. The patient characteristics of the 30 recruited are presented in Appendix 17

Table 15: Patient Characteristics

N	27
Age (mean yrs)	48 (range 19-65)
Gender, female (%)	22 (81%)
Education (mean yrs)	15 (range 11-20)
Insomnia subtype (n)	
Onset	9
Maintenance	8
Mixed	8
Early morning awakening	2
Insomnia duration (mean yrs)	18 (SD 17, 5 with childhood onset)

6.4.3.2 Treatment Outcomes for Sleep and Mood Variables

The sleep restriction intervention produced improvements in both sleep and mood variables, see results of the repeated measures ANOVAs in Table 16. Post-hoc Bonferroni corrected comparisons indicated significant improvements from baseline at post treatment and follow-up in insomnia severity (ISI), sleep quality (PSQI), dysfunctional beliefs and attitudes about sleep (DBAS) as well as anxiety (HADS-A) and depression symptoms (HADS-D). At both post-treatment and follow-up it took individuals significantly less time to fall asleep (SOL), and they

were awake less during the night (WASO) compared to baseline. In addition, early morning awakenings were significantly reduced at both timepoints compared to baseline. These improvements were associated with an increase in sleep efficiency (SE) and sleep quality (SQ) ratings at post-treatment and follow-up and total sleep time at follow up only. The effect sizes (Cohen's d(160)) for post-treatment and follow-up improvements were large for the four variables considered as outcome measures defined a priori: SE (posttreatment=1.7; follow-up=1.3), WASO (.9;.7), SOL (1.0;.7) and ISI (1.2;1.3). The baseline HADS-A score was in the mild region(546) despite the exclusion of individuals with anxiety disorders.

Table 16: Pre-treatment Sleep and Mood Characteristics with Post-treatment and Follow-up Changes

	Baseline mean (SD)	95% CI [lower;higher]	Post-treatment mean (SD)	95% CI [lower;higher]	Follow-up mean (SD)	95% CI [lower; higher]	F	DF	Sig.	Partial Eta^2
ISI	**19.2** (4.6)	17.4-21	**12** (7.6) ***	9-15	**11** (8.0) ***	7.9-14.2	**24.8**	**1.6; 40.6**	**<.001** [a]	**0.5**
PSQI Global	**13.2** (2.8)	12.1-14.3	**8.4** (3.8) ***	6.9-10	**7.7** (4.7) ***	5.9-9.6	**31.4**	**1.2; 33.1**	**<.001** [a]	**0.5**
DBAS-16 (/16)	**6.2** (1.7)	5.5-6.8	**5.1** (1.9) ***	4.3-5.8	**4.2** (2.2) ***$^{++}$	3.4-5.1	**31.4**	**1.5; 40.2**	**<.001** [a]	**0.5**
HADS-Anxiety (/21)	**9** (4.5)	7.2-10.8	**6.1** (4.9) ***	4.1-8.0	**6.5** (5.3) **	4.4-8.6	**13.7**	**1.6; 42.5**	**<.001** [a]	**0.3**
HADS-Depression (/21)	**6.1** (3-8)	4.7-7.4	**4.1** (1-8) **	2.8-5.6	**4.0** (1-6) **	2.5-5.4	**10.5**	**2; 52**	**<.001**	**0.3**
Sleep Diary										
TST (min)	**281** (98)	242.5-320.1	**304** (68)	277.5-331.4	**342** (90) **$^{+}$	306.2-378.1	**9.3**	**1.6; 41**	**<.01** [a]	**0.3**
SOL (min)	**82.6** (81)	50.3-114.9	**24.7** (30.5) **	12.7-36.8	**37.3** (57.6) **	14.5-60.1	**15**	**1.5; 39.6**	**<.001** [a]	**0.4**
WASO (min)	**59.7** (54.4)	38.1-81.2	**19.1** (29.9) **	7.2-30.8	**27.3** (33.1) *	14.2-40.4	**10.8**	**1.2; 32.8**	**<.01** [a]	**0.3**
EMA (min)	**64** (51)	43.6-84.6	**15** (16) ***	9.5-22.1	**19** (21) ***	11.3-28.6	**24.9**	**1.1; 30.5**	**<.001** [a]	**0.5**
SE (%)	**55.7** (18.8)	48.2-63.1	**83.5** (13.8) ***	78.0-88.9	**79.9** (18.8) ***	72.8-86.9	**42.1**	**1.6; 41.4**	**<.001** [a]	**0.6**
Sleep Quality (/4)	**1.3** (0.8)	1.0-1.6	**2.4** (0.2) ***	2.1-2.8	**2.4 (0.9)** ***	2.1-2.8	**32.9**	**1.6; 40**	**<.001** [a]	**0.6**

* significantly different from baseline at p<.05; ** significantly different from baseline at p<.01; ***significantly different from baseline at p<.001

$^{+}$ significantly different from post treatment at p<.05; $^{++}$ significantly different from post treatment at p<.01

[a] Greenhouse-Geisser corrections

ISI= Insomnia Severity Index; PSQI= Pittsburgh Sleep Quality Index, DBAS= Dysfunctional Beliefs and Attitudes about Sleep; HADS= Hospital Anxiety and Depression Scale, TST=Total Sleep Time, SOL= Sleep Onset Latency, WASO= Wake after sleep onset; EMA= Early morning awakening, SE= Sleep efficiency

6.4.3.3 Adherence

With an effective intervention established, the next paragraphs are concerned with the exploration of the adherence variables, beginning with descriptive information for the self-report questionnaire and the three methods (sleep diary, actigraphy marker, actigraphy movement). Thereafter, information is presented on possible differences across methods (sleep diary, marker, movement) for the composite score measurement. This section finishes with an analysis of the relationship between adherence and outcome.

6.4.3.3.1 Self-report Questionnaire

The measurement of adherence obtained from the questionnaire indicated that the majority of individuals reported following the bed restriction program "most" or "all of the time". With a possible range of 5-30 and higher scores indicating better adherence, total 4-week scores (sum of all items averaged over the 4-weeks) ranged between 11.5 and 30, with a median score of 26.3. Total average weekday, weekend, threshold and rising scores, over the 4-weeks, were also skewed; from a possible score of 6 (averaged responses across 2 items) median values were 5.4, 5.3, 5.3 and 5.1 respectively. Interestingly, there was a significant difference in self-reported weekday and weekend adherence rates; unsurprisingly, participants indicated they adhered to their bed/rise times more often on weekdays than weekends [Z = -2.6, p<.05]. There was no difference on self-reported adherence to threshold versus rising time (p=.69). Individuals were also asked to rate the extent to which they followed the recommendation during the follow-up period; similarly to the 4-week treatment ratings, individuals reported following the program, both in terms of overall adherence, as well as weekday, weekend, threshold and rising time items the "most of the time". Again, adherence on weekdays was significantly higher than on weekends [Z = -2.9, p<.01], but adherence to threshold and rising time were not significantly different (p=.21).

6.4.3.3.2 Sleep Diary

Based on the information provided in the participants sleep diaries, the median deviation of 7.6 minutes from the rising time [IQR= 2.2-24.5] was significantly

higher that the median deviation from the threshold time of 3.3 minutes [IQR= 3.4-9.6] from the threshold time, Z = -3.0, $p < .01$. The calculated adherence variables (composite score, rise and threshold deviation variance) that are obtained by the above values are presented in Table 17.

6.4.3.3.3 Actigraphy

6.4.3.3.3.1 Marker

At a descriptive level, information obtained from the marker revealed a median deviation of 13.5 minutes from the get up time [IQR= 6.3-26] and this was significantly higher that the deviation of 3.3 minutes from the threshold time [IQR= 0.6-17.3], Z = -3.3, $p < .01$. Adherence variables obtained from these absolute values are visually depicted in Table 17.

Table 17: Descriptive Statistics for Adherence measurements across methods

Adherence Variable	Sleep Diary*			Actigraphy *					
				Marker			Movement		
	Mean	SD	Median [IQR]	**Mean**	SD	Median [IQR]	**Mean**	SD	Median [IQR]
Composite Score	**17.7**	5.0	17 [14.5-20.3]	**19.1**	4.6	18 [15-23]	**21.7**	5.1	22.5 [16.6-25.8]
Rise deviation variance	**24.2**	25.4	15.3 [4.6-37.6]	**22.4**	21.5	17 [7-28]	**26.6**	19.9	27 [10.8-37.12]
Threshold deviation variance	**14.8**	19.2	9.7 [2.8-19.8]	**14.4**	20.2	1 [5.4-27]	**25.4**	17.0	27.7 [12.6-34.6]

*Higher scores indicate worse adherence

6.4.3.3.3.2 Movement

Analysis of the movement data also provided information about the extent to which individuals adhered to their agreed bed window. The median of the absolute deviation score of the average 28 days for the rising and threshold time was 21.5 [IQR= 4.4-38.1] and 13.6 [IQR= 3.6-30.4] minutes respectively. There was no difference between these times (p=.2). The mean and median values for each adherence variable are presented in Table 17.

6.4.3.4 Relationship between Adherence methods

A repeated-measures ANOVA comparing the three methods [sleep diary, marker, movement] across the composite score, revealed a significant main effect of method [$F(1.6;41.6)= 27.7$, $p<.001$; partial $eta^2=.5$, Greenhouse Geisser corrected] Post-hoc Bonferroni-corrected contrast showed significant differences between all pairwise comparisons. The composite score from both the movement and the marker data was significantly higher than the composite score obtained from the sleep diary. The movement composite score was also significantly higher from the same measurement obtained from the markers [all $p < .01$].

6.4.3.5 Adherence and Outcome

Treatment Outcome was classified in the follwing ways:

1. Sleep efficiency, WASO and SOL change score obtained from the diary.

2. ISI change score

The correlations between outcome variables and all adherence measures are outlined in Appendix 18. When adherence was assessed using the self-report questionnaire across the 4-weeks, there was no relationship between participants' ratings of implementation of the program and any outcome measures. Similarly, neither sleep diary nor actigraphy movement adherence variables were related to ISI change, or changes of sleep efficiency, sleep onset latency or wake after sleep onset. Moderate significant correlations were found between the composite scores obtained from the actigraphy markers and sleep efficiency change scores at posttreatment (*rho*=-.49; p=0.009) and follow-up (*rho*=-.40; p=.04). With higher scores on the adherence measures indicating

poorer adherence, these negative correlations indicate that greater adherence is associated with improvements in sleep efficiency, see Figure 13.

There were trends for a significant relationship between follow-up sleep efficiency and the following adherence measures:

- Sleep diary threshold deviation variance (*rho*=-.35; p=.066)

- Marker rise deviation variance (*rho*=-.38; p=.053)

It is important however, to interpret these results in light of the multiple comparisons conducted.

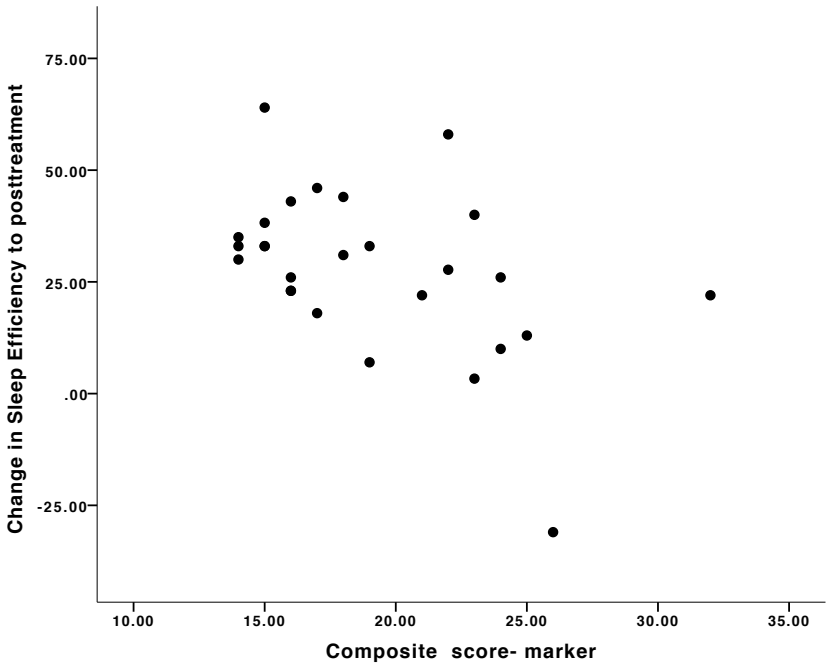


Figure 13: Relationship between composite score and post-treatment sleep efficiency change

In order to reduce the number of comparisons in subsequent analyses, the one adherence variable that was significantly correlated with outcome was selected for further testing:

1. Marker Composite Score

6.4.3.6 SOCSI Validity and Reliability

With both treatment effects and adherence measures established, this section focuses on the reliability and validity of the SOCSI. The first part of this section reports data on the stage of change component of the SOCSI, including frequency counts and stage transitions across the intervention weeks. The next part provides information on concurrent and predictive validity of the stage of change component. The self-efficacy component is subsequently examined in relation to both adherence and the stage of change component providing support for its validity. The section concludes with test-retest reliability analysis.

6.4.3.6.1 Stage of Change Across Treatment

Frequency counts for the Stage of Change across the baseline-post treatment and follow-up are plotted in Table 18. The majority of the sample (≈75%) scored in the action stage during weeks 1-4, there was considerably more variation at follow-up, perhaps indicating that some individuals relapse during the 2- month period between the last treatment session and follow-up. Thus, stage transitions across these timepoints were examined in addition to the stage allocations.

Table 18: Frequency Count for Stage at each treatment timepoint

	Baseline (n)	WEEK 1 (n)	WEEK 2 (n)	WEEK 3 (n)	WEEK 4 (n)	Follow-Up (n)
Pre-Contemplation	0	0	0	0	0	0
Contemplation	1	0	1	3	2	5
Preparation	26	7	5	4	3	7
Action	-	20	21	20	22	3
Maintenance	-	0	0	0	0	12
TOTAL:	27	27	27	27	27	27

6.4.3.6.2 Stage Transitions

Because stage of change was measured at each time point, it was possible to identify 5 types of stage transitions tracked across the 4-week intervention and follow-up period, these transitions are adapted from other descriptions.(547) At 4-weeks post-treatment, the majority (74%) had transitioned through the stages in a positive linear fashion- 'up through the stages'- the remaining individuals displayed a negative/non-linear transition. At follow-up, eight individuals had relapsed into a lower stage, moving from the positive linear category, to an inverted v-shaped pattern. Figure 14 is a visual representation of the five patterns along with percentages of the sample scoring in the respective categories at post-treatment (top pie chart) and follow-up (lower pie chart). Interestingly, those who transitioned in a linear fashion through till follow-up (from preparation to action stages) had better adherence (lower marker composite score) across the 4- week intervention phase than those progressing in a non-linear fashion, i.e. relapsing at some point, or not progressing; $U=49$, $z=-2.0$, $p<0.05$. There were no significant differences in adherence rates between stage transition patterns through till post-treatment.

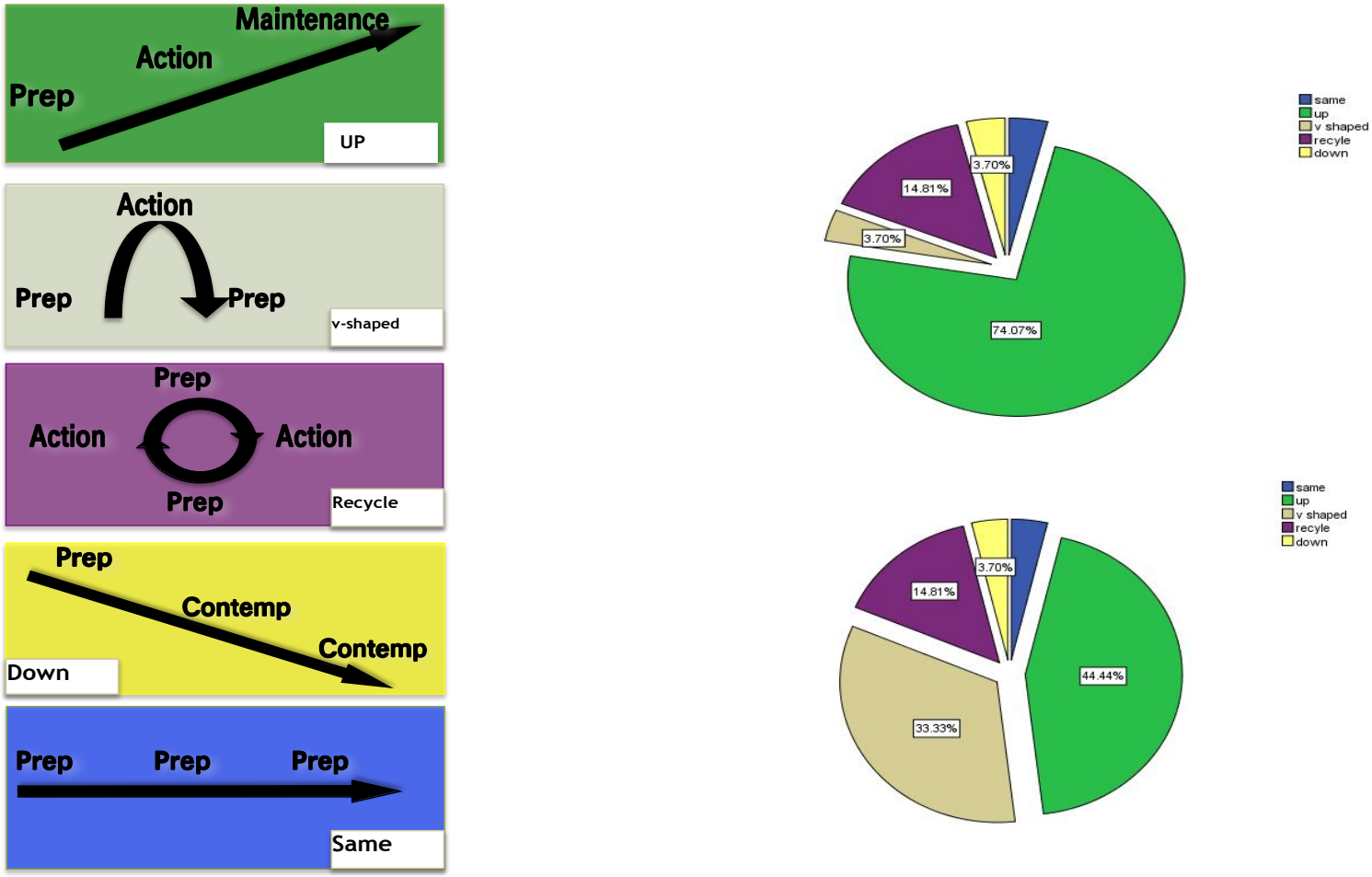


Figure 14: Stage Transitions
Stage Transitions (left boxes) across 4-weeks (top pie chart) and through till follow-up (lower pie chart)

6.4.3.6.3 Criterion-related Validity

To assess the validity of the stage of change construct in this sample, the relationship between stage allocation and a criterion was established. Adherence (the composite score from the marker) was identified as both a criterion for concurrent and predictive validity, and patient motivation was assessed at each time point to povide further evidence for concurrent validity. Baseline scores for stage of change were not included in this analysis because of a lack of variablility (see Table 18). The criterion variable was compared across those who scored in the preparation and action stage at week 1. For the remaining observation points (week 2-4 and follow-up), the contemplation and preparation stages were combined because of small frequencies counts in each stage.

6.4.3.6.3.1 Concurrent Validity

6.4.3.6.3.1.1 SOC and Adherence at each week

The relationship between stage of change and concurrent adherence was assessed for each observation point. Mann-Whitney tests revealed significant differences in adherence between stages at all weeks, as seen in Table 19.

Table 19: Differences in Marker Composite Scores between stages across the each observation time point

Stage of Change	Week 1	Week 2	Week 3	Week 4
	Median [IQR]	Median [IQR]	Median [IQR]	Median [IQR]
Contemplation/ Preparation	26 [20-30]	21 [18.8-27]	25 [18-28]	21 [19-30]
Action	15 [14-18]**	16.5[14.3-19.8]*	17 [15- 19]*	16 [14.8-22.3]*

Significantly different from adherence rates in contemplation/preparation stage at *p<.05; **p<.01

6.4.3.6.3.1.2 SOC and Motivation

When comparing stage of change with concurrently measured motivation, only the first week revealed significant differences, with those in the action stage scoring higher on the motivation scale [median=9.5; IQR=9.3-9.9], than those in the preparation stage [median=9; IQR=8.8-9.3], U=122, z=2.9, p<.01. There was a non-significant trend for week 3. Those in action stage [median=10.0; IQR=9.7-10.0] had a higher motivation than those in contemplation/ preparation stages [median=8.8; IQR=8.4-10.0], U=100, z=1.7, p=0.08. All other comparisons (week 2, 4; follow-up) were non-significant (p>0.05) see Appendix 19 for median values.

6.4.3.6.3.2 Predictive Validity

It was hypothesised that stage of change (SOC) measured at baseline would be related to adherence over the 4-week treatment period. With no variation in SOC at baseline-the majority (96%) of the sample in the preparation stage- week 1 SOC was used to predict adherence over the 4-week treatment period.

When measured at week 1, stage of change was related to 4-week adherence (marker composite score). Individuals who aligned themselves to the action stage were significantly more adherent across the 4-week treatment phase than those in the preparation stage. The composite score obtained from the marker was lower (better adherence) in the former group [median, 16.5; IQR= 15-19], compared to those in the preparation stage [median= 24; IQR=22-26], U= 20, z=-2.78., p<.01. Figure 15 shows a visual representation of this difference.

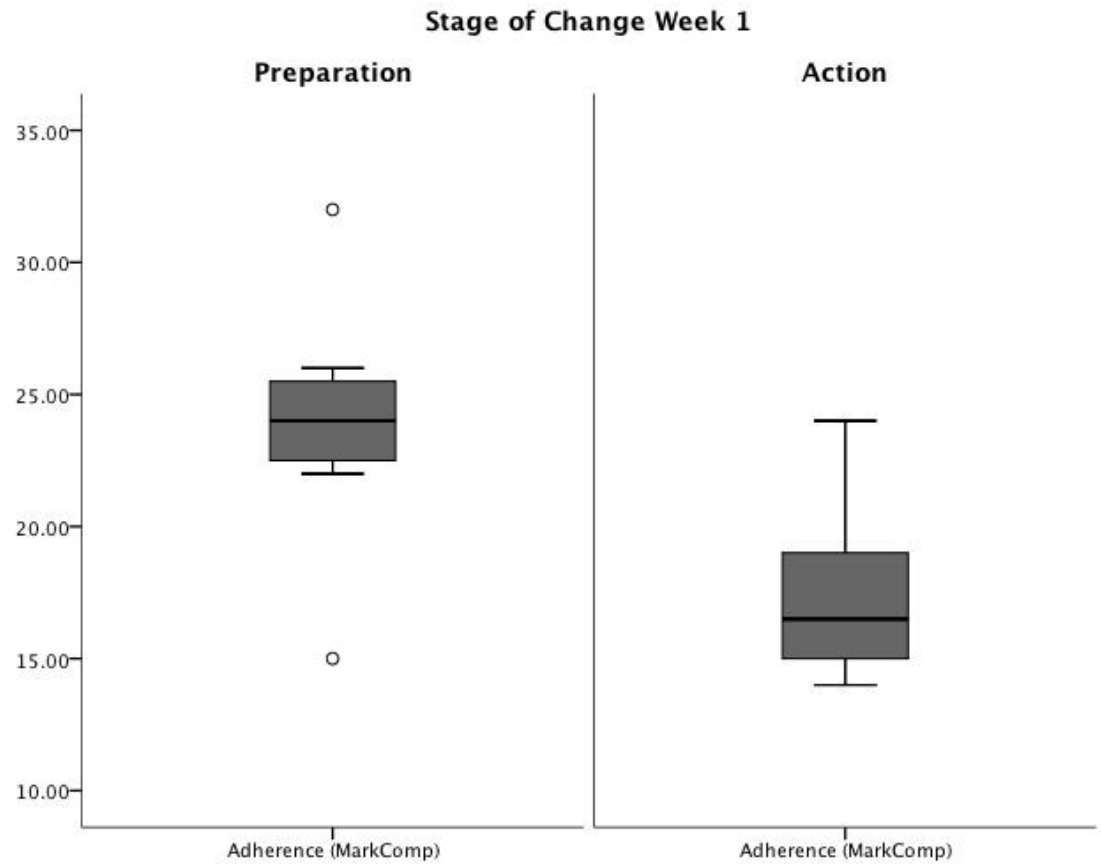


Figure 15: Difference in Adherence at week 4 between preparation and action stage (week 1)

6.4.3.6.4 Self-Efficacy

Individual's rating of their confidence in being able to follow the sleep restriction routine (self-efficacy), when measured before treatment was initiated (baseline), was moderately correlated with the composite score obtained from the marker, (rho= -.41, p<.05). When was measured one week into the intervention, the strengths of the associations increased slightly. Self-efficacy ratings at week 1 were significantly related to adherence (rho =-.46, p<.05).

The transtheoretical model predicts that individuals, who score in theoretically superior stages, will present with higher ratings of confidence in engaging in behaviour change [concurrent validity of the self-efficacy measure]. In this sample, there was a significant difference in self-efficacy ratings in those scoring in the action [median=4, IQR= 3.3-4] and preparation stage [median=3, IQR=3-3], when these were measured at week 1, U=112, z=2.7, p<0.01. Similarly significant were these comparisons at week 3 (p<0.01) and at follow-up (p<0.01). All other weeks were non significant (p>0.05).

6.4.3.6.5 Test-retest Reliability

Stage of change allocation and self-efficacy ratings were compared before and after the mental arithmetic test at week 1. The Intraclass correlation coefficient (ICC) for stage of change was 1, with all individuals scoring in the same stage. There was also a strong overlap for self-efficacy ratings; 26 of the 27 individuals reported identical self-efficacy ratings at both time points. One individual scored higher at time point 2; the ICC for self-efficacy was high accordingly (ICC=.95).

6.4.3.7 Content Analysis:

A total of 481 responses were provided from all 27 participants across the 6 observation time points. Of these, 10 responses (0.02%) were excluded from analysis. Seven responses to the disadvantages question were excluded because they were considered vague *('short term pain')*. Three responses to the processes of change question were not analysed because they did not qualify as a process of change ('can sleep consistently'). Of the remaining 471 analysable individual responses 107 responses were considered multiple units (e.g. 'short term fatigue, disruption to family life') and each unit was counted equal as described in the methods section.

6.4.3.7.1 Decisional Balance

6.4.3.7.1.1 Pros for adhering to the sleep restriction routine

The three most frequently generated advantages for adhering to the sleep restriction programme over all observation time points were:

1. Improvements in sleep quality/quantity (47.6%)

2. Improvements in energy/motivation (13.5%)

3. Improved outlook (7.4%)

Table 20 depicts the themes and relative frequency counts for data generated across all time points. The three most frequently reported themes are in bold.

Table 20: Themes and relative frequency counts: Pros for change

Theme	Example Quote	Relative Frequency Count (%)
Improved sleep quality/quantity	***Better quality of sleep and more sleep***	**47.6**
Improved Energy/motivation	***More rested , more energy to exercise and do things***	**13.5**
Improved Outlook	***Ability to make plans for holiday /social events***	**7.4**
Curing insomnia	*The treatment is working, so could lead to a cure*	6.1
Improved Emotional regulation	*Better mood*	5.2
Better sleep pattern	*Help get me into a good sleep routine*	4.8
Improved Performance at work/school/daily activities	*Able to arrange meetings*	3.5
Improved Cognition	*Better memory, concentration*	3.1
Less Concern about sleep	*Being more relaxed about sleep*	2.6
Improved Health/well being	*Improved health*	2.6
Improved Sleepiness	*More awake*	1.7
Treatment effectiveness	The evidence shows that this method is 70-80% effective	0.4
Improved Social functioning	*Better social life*	0.4
More time	*Better use of time*	0.4
Improved Appearance	*Have no bags under my eyes now*	0.4

The theme ‘improved sleep quality/quantity’ was most frequently cited as a pro at all time observation points. There was also stability in the frequency counts of the theme ‘improved energy/motivation’, with this the second most frequent pro at all time points, except for baseline (surpassed by ‘improved outlook’). There was considerably more variability in the third most frequently cited advantage across the weeks: the following themes ‘curing insomnia’, ‘better sleep pattern’, ‘health/wellbeing’ and ‘emotional regulation’ were rated the third highest advantages across different weeks.

6.4.3.7.1.2 Cons for adhering to the sleep restriction routine

Interestingly, of the total number of responses given across all observation points, 34.3% depicted no real disadvantage to implementation of the treatment regime. Following that, the disadvantages that *were* mentioned included ‘reduced energy/motivation’ (23.8%), ‘increased sleepiness’ (9.4%) and ‘disruption to routine and social activities’ (7.2%). Table 21 displays the themes along with representative quotes and respective frequency counts.

Table 21: Themes and Frequency Counts: Cons for change

Theme	Example Quote	Relative Frequency Count (%)
No disadvantages	***No real disadvantages***	**34.3**
Reduced Energy/motivation	***Tiredness during the working week***	**23.8**
Increased Sleepiness	***Short term problems with sleepiness***	**9.4**
Disruption to routine and social activities	***Restriction of normal evening activity***	**7.2**
Impaired performance at work/school/daily activities	*Reduced ability to work*	5.5
Disruption to family/household	*Keeping partner awake*	3.9
Migraines	*Headaches continuing*	3.3
Concern over loss of sleep/sleep opportunity	*Getting less sleep than I would like*	2.8
Impaired emotional regulation	*Continuing to feel worthless + irritable*	2.2
Fear of no change	*No change to insomnia*	2.2
Health/wellbeing	Not feeling so good	1.7
Need to find activities	*Finding things to do*	0.6
Lowered self-confidence	*Questioning my staying power (motivation)*	0.6
Social functioning	*Falling asleep at sociable hours*	0.6
Boredom	*Boredom in the evening*	0.6
Requires discipline	Requires discipline	0.6
Cognition	*Brain not functioning as well*	0.6
Burden	*Remembering to keep to restricted times*	0.6

Similar to the advantages, the two most frequently counted themes were stable across the observation points; except at baseline, where reduced energy/motivation was more often reported than no disadvantages. The third most frequently identified theme was variable across the time points: 'increased sleepiness', 'disruption to routine and social activities' and 'impaired performance at work/school/daily activities' represented approximately 10% of the responses made across different observation points.

6.4.3.7.2 Processes of Change

When asked to describe strategies that helped them implement the sleep restriction program, self-liberation strategies (43.3%) were most frequently quoted by individuals across all observation time points. This was followed by counter conditioning strategies (26.3%) and consciousness raising (16.0%), see Table 22.

Table 22: Themes and Frequency Counts: Processes of change

Theme	Example Quote	Relative Frequency Count (%)
Self-liberation	***Be more determined to improve***	**43.3**
Counter conditioning	***Find interesting things to keep me occupied***	**26.3**
Consciousness raising	***Think how bad I was before and what I can achieve***	**16.0**
Helping relationships	*Get support from sleep centre and friends who believe in bed restriction therapy*	9.3
Emotional arousal	*Think positively about overcoming my sleep problem*	2.1
Self-re evaluation	*Be confident to myself [sic]*	1.5
Stimulus control	*Try to limit things happening that bother me*	1.5
Cognitive processes		15.4
Behavioural processes		84.6

As described in Appendix 8, self-liberation strategies involve a process of making firm commitments for change, take responsibility and strengthen belief in own capabilities for change. Counter conditioning, an additional behavioural process, includes pursuing situations that facilitate behaviour change. In this sample, this process consisted largely of ways to find activities helping them to stay up until their threshold time. The cognitive strategy consciousness raising was a process that was also frequently raised, this process includes making use of informational resources- in this case to remember how bad the sleep was prior to treatment implementation and what improvements can be made- to raise awareness of the need to engage in the new behavioural pattern. Other behavioural strategies included helping relationships, i.e. getting support from significant others; and stimulus control- removing unwanted stimuli that would impede adherence to the sleep restriction program. Other cognitive processes included emotional arousal and self-re-evaluation, which reflected emotional thoughts about the intervention itself and in terms of personal values, respectively. At baseline counter conditioning was the most frequently reported strategy, closely followed by self-liberation. There was a crossover at week 1 that remained through till follow-up. Third highest frequency counts were exposed for consciousness raising across all weeks except for baseline and week two, where this strategy was surpassed and tied with helping relationships, respectively. Overall, behavioural strategies (84.6%) were more often quoted than cognitive processes (15.4%) and a similar ratio was present for all weeks.

6.4.3.8 Other predictors of Adherence

Exploratory correlation analysis was conducted to examine potential baseline predictors of adherence, in addition to the main results reported above. Of the possible baseline predictors of adherence, only the IPQ subscale 'illness coherence' was related to outcome. Interestingly, a Spearman's correlation revealed worse adherence (higher composite scores from the marker data) in those who had a greater personal understanding of their sleep problem rho=.50; $p<0.05$. There was a trend for the IPQ-R subscale emotional representation rho=.34; $p=0.08$. All other correlations (all other IPQ-R subscales, ESS and FSS) and were not significantly related to adherence ($p>0.05$), see Appendix 20.

6.5 Discussion

Adherence to medical treatments is less than optimal and the interest in improving adherence is steadily growing. Interventions with a theoretical underpinning have been considered more successful that their a-theoretical counterparts. One model specifically, the transtheoretical model of behaviour change, has gained considerable attention in the health psychology field, and its potential for understanding adherence to treatments for sleep disorders is emerging in recent investigations. The limitations of these studies translate into the need to:

- develop and cognitively pre-test a questionnaire to measure appropriately the components of the TTM model for the use in sleep research
- Determine the concurrent and predictive validity of the scale when applied to an insomnia population completing a bed restriction program
- examine additional subjective and objective measures of adherence to bed restriction therapy.

In order to achieve these goals, two studies were conducted.

6.5.1 Study 1

Study 1 describes the development and preliminary validation of the Stage of Change Questionnaire. The SOCSI was developed to meet the following criteria: assess all components of the transtheoretical model, to be a brief scale that can be used as both a research and clinical tool to assess and potentially modify stage of change and offer the opportunity, as a generic scale, to be adapted to other health domains. In order to accomplish these criteria, the questionnaire assessed stage of change and self-efficacy; and in addition recorded patient-generated pros and cons and processes of change. The latter feature, patient-generated responses from an open-box format, allows for the generation of qualitative data that can a) be used for further scale development to produce a *reliable* research tool that measures decisional balance and process of change but also used in its current format as a *valid* clinical tool that records **patient-specific** pros/cons and strategies for behaviour change. This process of content analysing patinet-generated reponses for providing potential items for the decisional balance has been utilised successfully in the marijuana use literature.(548) Using this open-response format for components of the

transtheoretical model, as a clinical tool, has not been previously reported. However, motivational interviewing, a technique that runs hand in hand with the transtheoretical model(359), describes the importance of patient-generated change talk in order to increase individuals' readiness to change and engage in treatment.(432) Lastly, the open response box for patients to record their 'action plan' discussed with their therapist, permits the possible adaptation of the SOCSI to other sleep disorders /health domains.

After the development of the SCOSI, the comprehensibility of the questionnaire was tested in a representative sample of the potential end users: individuals undergoing a CBT-I program. The cognitive pretesting highlighted issues with the questionnaire that need to be addressed before further reliability and validity assessment could be made. These change include minor issues with layout, confusing wording, and the use of jargon. More substantial changes were made to describe better the term behavior change of interest, i.e. 'the action plan'; distinguish better between the stage of change items preparation and contemplation, and use terms, such as 'advantages and disadvantages' and 'strategies to help stick to the action plan' that were easily understood for describing the decisional balance and processes of change components. Overall these changes to the SCOSI made it more comprehensible, were in line with the theoretical underpinnings of the TTM and were in concordance with the goals of producing a research and clinical tool to monitor and potentially enhance individuals' readiness to change. This latter point was particularly emphasised by three individuals remarking that completing the SCOSI led to a cognitive shift in their attitude towards the treatment. Because the majority of individuals in the cognitive pretesting sample preferred the algorithm version of the SOCSI, this was used in study 2 for further evaluation.

6.5.2 Study 2

Further validaty and reliability testing of the SOCSI occured in the context of a sleep restriction trial. Once prior assumptions had been met [1) the intervention produced significant effects and 2) this occurred as a function of adherence] the SOCSI showed to be a reliable and valid scale to measure elements of the Transtheoretical Model and their relationship to adherence.

In relation to the first assumption, significant treatment improvements were witnessed with the sleep restriction intervention. These outcomes are

similar to a trial with a similar study design;(139) although the baseline values in the present study are slightly lower for TST, WASO and sleep efficiency and higher for SOL, the effect sizes for improvements in the outcome measures SE, SOL, WASO and ISI at the follow-up were similar to those reported in Kyle et al.'s study.(139) The second assumption was also met. The composite score obtained from the actigraphy marker data was related to improvements in sleep efficiency at both post-treatment and follow-up. There were also non-significant trends for the variance in deviation from 1) the threshold obtained from the sleep diary 2) the rise time obtained from the marker. These correlations were moderate, but this is perhaps not suprising, considering the comparison of an objective (actigraphy adherence) and subjective (outcome) measure. The consistency/variance measures of non-adherence were not related to outcome in this chapter, although there were non-significant trends. The only associated measurements with outcome in Riedel and Lichstein's study [R&L], were adherence measures 'time in bed consistency' and 'rise time consistency'.(213) Intersetingly though, this was not replicated by others(208, 214). The adherence measures in this chapter were different from these previous studies, by considering any times within the agreed window (after threshold and before rising times) as adherent, which might explain differences in the results presented above. Whether consistency of bed/get up times [R&L] or variance in non-adherence [present study], is a stronger predictor of outcome needs to be evaluated further. There were also no relationships between self-reported adherence and outcome in this chapter. This contrasts with the Vincent et al. study(208), where self-reported adherence was significantly related to improvements in TST, WASO and insomnia severity. These correlations, similar to the ones reported in the present study, were however only moderate. Vincent et al.'s study presented bed restriction within a CBT-I package; any additional sleep-related benefits from other components could have had an effect on the the relationship between adherence and outcome and might explain these differences observed.

The main aim of the present study was to evaluate the validity and reliability of the SOCSI. To assess criterion-related validity of the SOCSI, the relationship between stage of change and concurrent adherence and motivation (concurrent validity) and future adherence (predictive validity) was examined. This approach to assessing validity of stage of change measures has previously

been reported in studies examining adherence(523, 549) and other helath behaviours(550-553). Results inidcated that adherence rates were poorer in individuals scoring in the contemplation/preaparation stage than in the action/maintenance stage. The motivation scale provided a further criterion the SOCSI was validated against. However, the only significant relationships was found during week 1, where scores on the motivation scale were higher in those who were in the action compared to individuals in the preparation stage. The failure in finding differences during the subsequent weeks, and only trends in week 3, might be a result of a ceiling effect, in the motivation scale, see Appendix 19; a finding that was also reported in the original paper.(531) Taken together though, this indicates evidence for the concurrent validity of this component; the inability to compare across all five stages, however reduces the confidence with which this claim can be made.

The relationship between stage of change at early phases of the treatment and later adherence provided information about predictive validity. Limited variability of stage of change at baseline prevented this time point from being used; however, at week 1, those individuals who scored in the action stage had better adherence across the 4-weeks intervention period, than those in the pre-action stage. This finding is consistent with the transtheoretical model and suggests that measurement of stage of change is helpful for identifying those who are more likely to adhere to the bed restriction intervention. To date this is the first investigation of stage of change in respect to adherence to bed restriction. Only two studies in the insomnia literature have investigated components of the transtheoretical model. Belleville and colleagues reported that none of the TTM components at baseline were related to subsequent hypnotic tapering, yet when self-efficacy when measured at a later stage, this variable was related to hypnotic use.(511) Perhaps more comparable to the SOCSI study design was an investigation of the TTM model for predicting adherence to an internet-based CBT-I.(223). In this study, individuals who were more contemplative had better self-reported adherence to sleep hygiene rules. No relationship was found between readiness to change and sleep restriction. This might seem contradictory to the results of the present study, however the concept readiness to change as assessed in Hebert et al.'s study is a more generic concept, which represents people's motivation to change(513), as

opposed to stage of change, which represents the stage individuals are at in the change process.

As integral aspects of the transthoeretical model, stage transitions and self-efficacy were examined in addition to stage of change. Although the majority of individuals transitioned through the treatment in a linear fashion, 30 % of the sample relapsed from the action stage at post treatment to the preparation stage at follow-up. This potentially identifies a critical period during which individuals might need further support, possibly with booster session, to ensure continued adherence to the bed restriction routine during this follow-up period. Of course 'falling' back into the preparation stage does not necessarily indicate a relapse; individuals might have improved to the extent they do not feel the need to continue with the routine.

Self-efficacy measured at baseline was related to adherence and this relationship increased when self-efficacy was measured at week 1. The increased strength in relationship between self-efficacy and stage of change throughout the weeks of treatment mirrors previous results reported in this area.(231, 286, 511) The relationship between self-efficacy and stage of change is an integral part of the TTM; this is somewhat supported with the results in the present study reporting higher self-efficacy rates in the action compared to the pre-action stages at week 1, 3 and follow-up. This also provides further information about concurrent validity of the stage of change component. Why there were no differences in the other weeks is however suprising.

In summary, these results indicated good construct validity of the SOCSI and contributes to the evidence supporting the use of TTM construct stage of change and self-efficacy as useful in understanding behaviour change in this population.

Test-retest of the stage of change and self-efficacy components revealed very high correlation coefficient. This suggests the SOCSI is highly reliable in measuring stage of change and self-efficacy. However as noted above, test-retest reliability is not paramount in assessing scales measuring components of the transtheoretical model. The choice of such a short timeframe is controversial, however this was deemed most appropriate, considering the theoretical underpinnings of the model. The magnitude of overlap between the two timepoints reported here is slightly higher than a study examining test-retest in a similarly short time span. Donovan's et al. show pearson correlations

of around .78 .72 and .59 in individuals who wanted to reduce smoking, reduce alcohol intake and increase exercise respectively. The use of the pearson's correlations were validated here with high frequency counts in all stages. The considerably larger sample sizes (n=404 of their interviewed sample wanted to reduce smoking, n=57 reduce alcohol intake and n=704 inrease exercise) and different population of interest, might account for these differences in magnitude of relationship to that reported in the present study.

Because no validated scales currently exist for decisional balance and processes of change for adherence to CBT-I, open response boxes were used to generate individual's own responses. Content analysis of these entries indicated considerable stability across the pros, cons and processes generated each week. There were changes in the highest frequency generated themes from baseline to week 1 and this might be an artefact of comparing responses before and after treatment initation. Important advantages for change seem to be improvement in nightime symptoms (better sleep quality, quantity and pattern), daytime symptoms (energy/motivation, , emotional regulation, cognition, performance etc.), as well as more global changes (outlook, curing insomnia, appearance, having more time etc.). Overall the sample most often reported no real disadvantages for adherence. This finding is potentially linked to the other results reported in this sample: overall good adherence to the bed restriction therapy and the majority of individuals scoring in the action stage, with limited spread across other stages. So, although it was not possible to create a decisional **balance** and examine its relationship with the stages of change, this finding provides partial support for the TTM, which claims that individuals moving towards the action stage will find the pros outweigh the cons for change. Disadvantages that were noted included impariments in daytime functioning (energy/motivation, sleepiness, performance), changes to personal/family life (disruptions to social activities, family life) and a general diffiuclty in adhering to the bed restriction (boredom, requiring discipline). This finding corroborates previous reports that adherence to bed restriction is not as straight forward as assumed, and may incur unwanted effects that may impact on continued implementation of the routine.(139). Overall, participants generated more behavioural (e.g. self-liberation, counter conditioning) than cognitive processes (e.g. consciousness raising, emotional arousal). The TTM proposes this kind of pattern is representative of transition through later stages (action through to

maintenance). Because a large percentage of individuals resided in the action stage throughout the treatment, these results might offer preliminary support for the processes of change in this population. Although only cautious conclusions can be drawn from these generated themes, this information does provide important material that can be used in the further development of decisional balance and processes of change items.

A secondary aim of study 2 was to evaluate further subjective and objective adherence measures. Four different methods were used for this: self-report questionnaire, sleep diary, and actigraphy marker and actigraphy movement data. Results from the questionnaire highlighted that individual's self-reported high rates of adherence; with a possible total score of 30, more than half of the sample scored over 26. These pattern of extreme skewness is similarly reported in other studies. Vincent and colleages(208) sample indicated, also on a scale from 5-30, a mean self-reported adherence of 21.4 (SD= 4.4). In a randomised control trial comparing CBT-I with zopiclone or placebo, Sivertsen et al.(210) reported individuals in the CBT-I arm had an average self-reported adherence of 4.8 [SD, 0.1] based on a 5-point scale. With these highly skewed rates is it perhaps unsuprising that relationships between self-reported adherence and outcome were not reported in the SOCSI study.

In addition to self-report, the SOCSI study made use of sleep diary and actigraphy (marker and movement) as measures of adherence. The absolute deviations obtained from the sleep diaries in study 2 are somewhat comparable to previous studies using sleep diaries. The individuals in the present study, according to their diary entry, went to bed on average 7 minutes before their threshold time and got up 19 minutes after their rising time across the 4-weeks intervention period [of note, the results section reported the median values, as these variables were not normally distributed]. Riedel and Lichtstein (R&L) report that individuals spent on average 27.89 minutes more in bed than agreed. Other studies have used or adapted R&L's measurements of adherence.(162, 208, 214) Vincent et al. 2003 calculate time in bed and wake (rather than rise) time variance, however fail to report any absolute values of minute deviation and in their 2008 study they only report wake up time variance. In a study combining CBT-I with mindfulness for individuals with psychophysiological insomnia, Ong et al. 2008, report the deviation of time in bed from agreed time was 21.95 minutes (SD=38.32 minutes). Additionally, individuals rose 30.46

minutes (SD=36.42 minutes) later than their agreed rising time. It seems that the degree of non-adherence (in terms of absolute deviation) to the bed restriction in this study is within the same range as reported by R&L and Ong's studies, although it is important to note that in the SOCSI study, time points before the threshold time and after get up time were set to a value of zero, thus values reported here might be slightly higher than if calculated in the way R&L and others have done. Thus, comparisons here are to be interpreted with caution.

The three measures used to evaluate adherence are very different to the ones used in previous studies. The composite score was a more conservative measure of absolute deviation from threshold and rising time with each of the first three 15 minute gradiantes being reduced to a simple scale of 1-3. The remaining two measurements reflected the variance in non-adherence, by calculating the variance of absolute deviation from both the threshold and rising time. To date, no one has computed a composite score in this way and the two consistency measurements are different from R&L's reported consistency, whereby the variance in actual time in bed/get up time is calculated. To cacluate their measures, R&L included timepoints **after** the threshold time and **before** the rising time. This of course is valid when examining consistency of time in bed/get up time per se, and the importance of consistency in bedtimes is not disputed; however with the objective of measuing adherence to a bed restriction program, where individuals are instructed to go to bed at **or after** a specific time (as done in the present study), including these times becomes less appropriate. In the present study, by setting threshold and get up times within the agreed window to zero, results showed that time deviation variance was 24 minutes, and threshold deviation variance was 15 minutes. Based on these differences, comparisons with R&L's consistency measures is not possible. A further difference to R&L's study is the intervention: sleep compression. Rise time consistency, was thus computed from the difference in consistency at baseline to consistency at post treatment. Whereas the present study allowed for consistency calculations throughout the treatment process. Thus, difference in treatment protocols also make comparisons difficult. Ong et al.'s study is perhaps more comparable to the SOCSI study design, however they do not report a measure of consistency.

In addition to sleep diaries, the present study also made use of actigraphy to obtain a measure of adherence and this method presents an interesting

comparison to the previously mentioned sleep diary. When comparing the three adherence methods (sleep diary, marker and movement data) for the composite score measurement, there were significant difference at all levels. It is difficult to determine which of these methods is the most objective, however the results of the ANOVA indicated that individuals are most adherent, when measured with the sleep diary, followed by the marker and lastly actigraphy movement. This partly supports findings from Carney and colleagues, who reported that a group of good sleepers, who were asked to restrict their time in bed across 2 nights and were unaware that the actiwatch would record their adherence, had significant discrepancy between their actigraphy bed time and agreed bed time, no discrepancies were reported in the sleep diary adherence measures. Although not reported, this might be synonomous with a significant difference in adherence across these two measures, similar to the comparisons in the present study. The group who was made aware that the actiwatch recorded adherence followed the routine better, both in actiwatch and sleep diary recordings. Participants in the present study were not told the actigraph would measure their adherence, however a small number of individuals did remark that the device would be able to determine whether they were “lying or not”.

Despite these considerations, all methods (sleep diary and actigraphy) reveal that individuals are very adherent to the bed and rising time (within about 30 minutes), and are better able at following the threshold time compared to the rising time recommendations, as well as on weekdays compared to weekends (self-report). Whether 30 minutes is an adequate cut-off for optimal adherence to bed restriction treatment seems possible, however needs to be further investigated.

Baseline variables (sleepiness, fatigue, dysfunctional beliefs and illness representations) were examined as possible predictors of subsequent adherence in exploratory analysis. Contrary to expectations, only one variable was predictive of adherence, sleep problem coherence. Interestingly the relationship was possitive, indicating greater personal understanding of the sleep problem at baseline was predictive of non-adherence to the bed restriction therapy. This contradicts assumtions of the original self-regulatory model, which would propose less coherence of the problem to be associated with poorer adherence. One potential explanation could be that individuals struggling to make sense of their sleep problem were more motivated to engage in the treatment. However,

as with the results above, the correlations are only moderate, and because of multiple comparisons made, these have to be interpreted with caution.

In summary, the results of study 1 and 2 have contributed to accomplishing the aims outlined for this chapter:

- Develop and cognitively pre-test a questionnaire to measure appropriately the components of the TTM for the use in sleep research (STUDY 1)

- Determine the concurrent and predictive validity and reliability of the scale when applied to an insomnia population completing a sleep restriction program (STUDY 2)

- Develop further subjective and objective measures of adherence to CBT-I (STUDY 2)

It is important to consider these results in light of the limitations of the studies.

6.5.3 Limitations and future directions:

The first limitation concerns the sample in study 2. Partly as a result of small sample sizes and highly motived research participants, there were not enough people in each stage of change, forcing the merge of the contemplation and preparation stage. Merging contemplation and preparation into one stage and contrasting adherence rates to those in the action stage is akin to predicting furture from past behaviour (the qualitative difference between action and preparation/contemplation is by definition the presense of behaviour change, i.e., adherence). With the merge, the quality of stage differences is lost and therefore the complete use of the transtheoretical model cannot be determined. Future studies determining the validity and reliability of the SOCSI will need to use larger sample sizes and investigate stage of change in a clinical, rather than a highly motivated research sample, to ensure a large distribution across all individual stages. Individuals with co-morbid psychological or medical problems and/or taking medication for their insomnia might be less ready for change and thus more likely to reside in the contemplation or pre-contemplation stage. Secondly, with the lack of valid decisional balance and processes of change measures for CBT-I adherence, open box response format was used. Thus the

reliability and (criterion-related) validity of these two measures could not be investigated. This study was only able to draw conclusions on the type of pros and cons conisdered important in this sample, and could not make inferences on how the **balance** between these two might relate to adherence. Although results of study 1 provided evidence for the face validity of these measures, there is a need to construct items using the results from the content analysis and develop scales for the decisional balance and processes of change that can undergo further validity and reliability testing.

Study 1 indicated good face validity of the SOCSI. Three individuals remarked (unprompted) on the ability of the SOCSI to make them aware of their motivation towards behaviour change. However, this might be problematic for the interpretation of study 2, where individuals could have been aware of the assessed construct of the SOCSI and responded according to demand characteristics. The potential of individuals becoming aware of the rationale for using the actiwatches to monitor adherence might have increased this effect even further.(see 228). Additionally, the mere completion of the SOCSI might have had an influence on adherence itself (both positive and negative). Generating patient-specific information on the pros/cons and processes of behaviour change might have made people more aware of their beliefs and attitudes towards treatment.

Using an actiwatch as a measure of objective adherence certainly furthers our understanding of adherence to a bed restriction program. Although the Respironics software determined rest periods have been validated against sleep diaries(554), the automatic scoring is corroborated with visual examination of the data, which may introduce an element of subjectivity and error. Furthermore, the participants in this study were undergoing sleep restriction therapy and potentially spending time resting prior to their threshold time (e.g. on the couch watching T.V.). This extended amount of inactivity (or lowered activity) compared to an active good sleeper retiring to bed at 10, might have led to an overestimation in the minute deviation for the threshold time in the movement compared to the sleep diary and marker data; the results of the ANOVA might support this possibility. Thus, the further development of ways of obtaining an **objective** account of adherence to the behavioural components of CBT-I is warranted. Potentially, the ananlysis of in-lab studies with concurrently measured actigraphy, sleep diary entries and video recording might shed light on

this issue. This will hopefully lead to a clearer understanding of what actually constitutes adherence to bed restriction therapy: is it sticking to the threshold/rise time to the minute? How much variation would be considered acceptable or even in fact normal error? These questions are currently left unanswered. Perhaps even more crucial, what is the individual's own definition of adherence and does influence the response to the SOCSI items? Certainly in the cognitive pre-testing study no clear differences in the definition of the term 'sticking to the action plan' emerged across participants. However, the clear identification of the behaviour of interest is vital in order to distinguish pre-action from action stages(503) The term 'consistently' was added to the questionnaire to standardise the potential definition of adherence as much as possible.

Hence, further clarification of optimal adherence to bed restriction therapy would be fruitful. This could also be accomplished by exploring various 'doses' of adherence to establish the relationship between adherence and outcome. The randomisation of participants to a bed window extended by 0, 15, 30, 60, 90 etc. min to artificially mimic varying degrees of non-adherence could provide information for establishing a consensus of optimal adherence, mirroring the efforts in the CPAP literature.(see 57)

The use of the transtheoretical model in understanding and predicting adherence to bed restriction is outlined above, however there are important alleged limitations of the model itself, that warrant consideration. The transtheoretical model was initially conceptualised to understand smoking cessation. This behaviour is relatively simple and inferences about self-reported stages of change are easily made. However, bed restriction is a complex treatment, involving the implementation of several behavioural changes: going to bed at or after the threshold time, getting out of bed at rising time, no napping. Thus the application of this model to more complex behaviours might be problematic at a conceptual level. A recent commentary described this as a potential explanation for the limited evidence for TTM guided stage-based interventions in improving activity levels.(555) Another alleged criticism of the model is the stage allocation process itself. Many have argued that the stages describe arbitrary cut-offs (e.g. 6 months is often used as the cut-off between action and maintenance). However the authors state this process is akin to the use of cut-offs in the medical field, e.g. to quantify the severity of a disease;

and that the value of the TTM is the inclusion of these discrete variables along side continuous ones, i.e. self-efficacy, decisional balance and process of change.(556) Increased use of the SOCSI and research into adherence to bed restriction therapy is needed, to ensure the cut-offs for stage allocation used in the SOCSI are valid.

A further criticism of the TTM is that stages are descriptive, not predictive, and that stage progression is not reflective of actual behaviour change.(557) The authors of the TTM respond to this criticism with studies indicating good predictive validity of the stages and limitations of studies that do not.(556) This present study certainly provides some evidence that the stage of change is predictive of behaviour change: i.e. adherence to bed restriction.

Lastly, some have argued that staging implies individuals make 'coherent and stable plans' and possible response options do not inclusively reflect their thought processes.(see 557). This might be true at the moment-by-moment level, where other processes, e.g., in this population, experienced levels of sleepiness, potentially influence their decisions. However at the global level, the cognitive pre-testing of the SOCSI was one way of establishing whether the thought processes of these individuals matched their response patterns, which, arguably to the subjectivity of the interviewer, was accomplished.

With the hope of addressing at least some of these issues with future studies, there is potential for the SOCSI to be implemented into routine clinical care of individuals with insomnia. The early identification of patients in the pre-contemplation or contemplation stage, who are less likely to adhere to the behavioural components of CBT-I and might benefit from increased cognitive restructuring before implementing the behavioural strategies, seems fruitful. In contrast, those in the action stage could be introduced to behavioural components immediately, being offered cognitive strategies later if needed. Thus stage matching based on the TTM model in the insomnia population warrants investigation, see also chapter 4. Some have argued that these types of stage-matched interventions in improving health related behaviour are not considerably successful(558); however the original authors state that studies providing evidence against stage-matching often fail to consider all components of the TTM model.(556) Previous efforts to tailor CBT-I treatment to individuals with different subtypes of insomnia have not been effective,(474, 475) however the nature of insomnia as a psychobiological disorder, which is optimally treated

with **cognitive** and **behavioural** techniques, certainly lends itself to tailoring the CBT-I treatment to stage allocation. An alternative to this matched approach might be integrating aspects of motivational interviewing(432) into the CBT-I program. This technique is currently described as 'a directive, client-centred counselling style for eliciting behaviour change by helping clients to explore and resolve ambivalence.'(433, p.326) If individuals are experiencing ambivalence when asked to adhere to the behavioural and cognitive components of CBT-I, motivational interviewing might help resolve this ambivalence before the individual engages in the behaviour change. This technique has recently shown to be successful in improving CPAP adherence.(354, 355, 358).

Lastly, it might be useful to match pharmacological versus behavioural techniques to stage allocation. Those in the pre-contemplation/contemplation stage might benefit first from short-term improvements with pharmacological options before being introduced with a more cognitive-behavioural option, once progressed through to a preparation/action stage. This would certainly add to the recent efforts to improve the effectiveness of insomnia treatment by combining these pharmacological and cognitive behavioural techniques.(166, 167)

6.5.4 Concluding remarks

This chapter presents the development and psychometric evaluation of the Stage of Change Scale Specific for Insomnia. Results suggest that the SOCSI is valid and comprehensible to individuals. The brevity and easy of administering the SOCSI supports its potential for being an integral part of the therapeutic process in daily clinical practice. Further psychometric testing of the SOCSI indicated that documenting stage of change and self-efficacy at early periods during therapy can provide useful information about who will adhere to a bed restriction program. The chapter also highlighted the use actigraphy in assessing adherence to this type of behavioural treatment for insomnia. Future studies need to assess stage of change in larger samples including clinical populations (e.g. with comorbid medical and psychological disorders and/or concurrently taking hypnotics), and investigate adherence to bed restriction, to permit the full evaluation of the transtheoretical model and its applicability in this population.

Chapter 7- Overarching discussion

7.1 Summary of the work conducted in relation to thesis aims

The introductory chapter to this thesis presented the literature on adherence to treatments for the two most prevalent disorders of sleep and wakefulness: CPAP for obstructive sleep apnoea and CBT-I for insomnia. Despite rigorous evaluations of the efficacy of these treatments, no single predictor has been consistently linked to CPAP adherence. The literature on CBT-I is very much under researched, with limited information on optimal rates/cut-offs for adherence or the relationship between adherence and outcome. Thus both areas are currently at the cusp of an agenda change. The insomnia literature requires a consensus on how to measure and conceptualise adherence to components of CBT-I. This progress will hopefully mirror the pathway the CPAP literature has taken. The further advanced CPAP literature, on the other hand, might benefit from paralleling recent efforts in other domains of health and adherence research. As outlined in chapter 1 and 2, the CPAP literature has largely become dichotomised. A holistic approach, integrating biomedical, psychological and social variables into a biopsychosocial model of CPAP adherence is needed. As such, three core targets for CPAP research were derived from the conclusions of the narrative review in chapter 2. Firstly there is a need for an extensive investigation of the largely under researched psychological and social variables that might influence CPAP adherence (Aim 1). Secondly, biomedical and psychosocial factors need to be considered as interacting variables influencing CPAP use; not only in a multifactorial, but also holistic fashion (Aim 2). Lastly, to translate this biopsychosocial approach into routine clinical practice, brief scales with good validity (not least face validity) measuring psychological and social variables need to be developed and rigorously tested, adding to already established biomedical measures (Aim 3). These conclusions were primarily drawn with CPAP adherence in mind, where the divergence between biomedical and psychosocial approaches was most evident. However, these conclusions can be equally extended to the CBT-I literature, or even to any area of adherence

research; ensuring adequate adherence is a health care task and after all, this, according to Engel, is managed best within a biopsychosocial framework.

To contribute towards these aims, four studies were completed within the remit of this thesis. Table 23 summarises the main outcomes of each of the four studies included in this thesis.

Table 23: Summary of main outcomes by study

Experimental Chapters	Aim of study	Methodology used	Summary of results	Thesis aim targeted
Chapter 3- A longitudinal qualitative analysis of the experience of using Continuous Positive Airway Pressure	Investigate the experience of CPAP adherence to understand further psychosocial variables predictive of machine use	Longitudinal approach, interviewing OSA patients before CPAP initiation, 1 week and 3 months after machine purchase using semi-structured interviews	CPAP embodies a treatment some individuals are resistant to use. Many resolve the conflict between feeling abnormal and longing for normalcy, begin to integrate CPAP into their life, making use of internal and external resources and motivators.	1
Chapter 4: A thematic analysis of patient experience of cognitive behaviour therapy for insomnia (CBT-I)	Understand how individuals experience the implementation of CBT-I components using a qualitative approach	Semi-structured interviews with individuals with insomnia who had recently completed a CBT-I program for their sleep problem	Establishing the meaning of CBT-I at the global level is fundamental. Individuals then begin to focus on the components, evaluating each component against certain criteria. CBT-I remains a challenging treatment with associated obstacles that need to be overcome.	1
Chapter 5: The effect of continuous positive airway pressure usage on sleepiness in obstructive sleep apnoea: real effects or expectation of benefit?	To examine the extent to which physiological and psychological effects account for the relationship between CPAP adherence and outcome	Patient-level meta analysis of three randomised placebo controlled cross-over trials identifying the effects of CPAP on daytime sleepiness	Above physiological effects, some of the outcomes witnessed from using real CPAP are a result of psychological effects, potentially influenced by an expectation of benefit	2
Chapter 6: The Stage of Change Scale for Insomnia (SOCSI) - A new scale to monitor stage of change during sleep restriction therapy for insomnia	Develop and validate a scale to assess components of the transtheoretical model (stage of change, self-efficacy, decisional balance and strategies for change)	Cognitive pretesting for the development of the scale (study 1) and validity and reliability testing in the context of a sleep restriction trial (study 2)	Good face, concurrent and predictive validity was established for the SOCSI. The scale provides an optimal measure for assessing stage of change to understand adherence to sleep restriction therapy	3

The first two experimental chapters (3 & 4) used a qualitative methodology to allow for a detailed and thorough investigation of individual's thought processes whilst implementing treatment recommendations for their sleep problem. This method provides a window into the patients' attitudes and beliefs and enables the identification of psychosocial variables that need to be considered in interaction with biomedical variables (Aim 1).

As summarised in Table 23, the results of chapter 3 confirmed that CPAP is perceived as an intrusive therapy and revealed how individuals battle with the conflict between the perceived need for treatment and the simple unwillingness

to use the machine. Longing for normalcy highlighted the dichotomy between feeling abnormal with the machine and the desire to regain normal functioning during the day. These resolved in some, however remained present for others despite the attempts to integrate CPAP into their life, making use of motivators and resources. Reflecting on these results in light of the aims of the thesis several points can be mentioned.

Both self-endorsed and externally inflicted stigma contributed to the unwillingness to accept and adhere to treatment. Feeling old, sick or lazy were common attributes of this stigma. Although emerging in some of the qualitative studies, this has not been quantitatively explored and its contribution to adherence within the biopsychosocial model needs to be assessed. For example men might be more inclined to endorse the feelings of stigma compared to women. The acceptance of the sleep disorder and adherence to treatment might be equated with taking on a sick role and this may stand in opposition to the authoritative/protective role some men might still take on within the family. Age might also be differentially associated with stigma. As outlined in chapter 1, some studies have indicated younger age to be associated with poorer adherence. The perception of needing “oxygen therapy” to help one to sleep and to breathe is equated with feeling old, and this may result in resistance in younger individuals. Lastly, this psychological variable might interact with social factors, such marital status. The contrast between taking care of one’s family and taking on a sick role, is less relevant for single (male) individuals. Turning the attention to other health domains and adherence research, a well-validated scale outside the CPAP literature is the Illness Perception Questionnaire-Revised (IPQ-R)(533) and its subscale emotional representation, which assesses the extent to which negative emotional connotations are attached to the illness; an example item of this subscale is ”I get depressed when I think about my sleep problem”. This scale might tap into the construct of stigma, however does not depict an exact representation of it. Nevertheless, the IPQ might prove useful in understanding the emotional representations these individuals might endorse.

A further aspect emerging from the results of chapter 3 that has been largely neglected in the CPAP adherence field is the impact of social factors. As outlined in chapter 1 and the discussion of chapter 3, variables such as spousal support and improvement in partner’s sleep quality have been associated with CPAP adherence. However, the results of this qualitative investigation highlight

the need to expand our understanding of how both the illness and treatment impact on the patient's surrounding. The individuals in this sample expressed concerns about how the integration of CPAP would affect their bed-partner, but at the same time were encouraged to adhere to treatment as a result of spousal support and the potential improvements in their partner's sleep and their relationship. These results emphasise the importance of involving the partner into the clinical setting, as has successfully been done in interventions to improve CPAP use(e.g., 351). The consideration of the patient's social network also coincides with the merits of the biopsychosocial model- seeing the patient from a holistic perspective. Consequently, not only must further exploration of social variables occur, but also effort should be made to understand how these factors relate to variables within the biomedical and psychological domain. For example, extensive partner support might be useful in dealing with adverse effects or support might be beneficial in those with high, but not those with low disease severity, as has previously been shown.(338) Thus chapter 3 highlights additional psychological and social variables that could be considered in relation to CPAP adherence, and lays a path for the investigation of possible interactions, built on the recent efforts described in chapter 2.

As a result of the dearth of information on CBT-I adherence and limited qualitative exploration in this area, the aim of chapter 4 was to offer an in-depth account of the individual's experience, in order to expand our, as of yet, limited knowledge on adherence to CBT-I. Similar to chapter 3, the results of this qualitative investigation pointed towards the importance of considering social aspects. This was prominent in the subthemes "evaluation of self against others" and "negative/unwanted consequences". These issues pertain to two important issues that need to be mentioned here. Firstly, the delivery of CBT-I in a group format, and secondly, the role of family/bed partner in the cognitive behavioural treatment of insomnia. Two studies have established a group format to be as equally effective as individual CBT-I delivery in head-to-head evaluations,(477, 478) however neither reported on potential differential effects on adherence. From the results in chapter 4, it is evident that the group format had an effect on the individuals' behaviour. The investigation of the intricate relationship between group dynamics and adherence to CBT-I in quantitative studies is warranted.

The influence of the bed partner has largely been neglected in the understanding of the onset, maintenance and treatment of insomnia, as outlined in a very recent literature review.(559) Chapter 4 results indicated how individuals are concerned that the implementation of some behavioural components will impact on the bed partner and household in general. In this recent review, Rogojanski and colleagues outline the potential beneficial effect of integrating the bed partner into the treatment environment, mainly to encourage support with adherence, but also to educate the partner about behaviour that may be maintaining the problem, including those of the partner. The potential impact behavioural components may have on the partner's sleep are unfortunately not addressed in this review, despite the negative consequences this could have on augmented support. The quantitative exploration of this relationship may be fruitful; as well as addressing the partner's sleep, or at least the patients concern's about negative consequences, within the clinical setting. In summary, the results of chapter 4 highlight the need to provide fundamental information about factors within the social domain of a BPS model contributing to adherence to CBT-I. Chapter 4 also highlights the dynamic nature of adherence to CBT-I, within the context of two themes: "ongoing evaluation of components" and the subtheme "is the timing right", indicating that the psychological construct readiness to change might be valuable in understanding treatment adherence (as further evaluated in chapter 6). In respect to biomedical factors, side effects associated with implementation of behavioural components were identified in this qualitative investigation. This has previously been established in a trial on sleep restriction,(139) however has not been identified in a study of multi-component treatment. The further quantification of how side effects may play a role in patient adherence is warranted, as are possible interactions with psychological and social variables. For example, increased daytime sleepiness and reduced functioning as an unwanted side effect of sleep restriction might fuel the dysfunctional belief of requiring 8 hours of sleep in order to ensure adequate next day functioning. In summary, chapter 4 highlights potential psychological, social and biomedical variables that yet have to be explored within the framework of a BPS model.

The two qualitative chapters were explicitly designed to expose possible psychological and social factors that predict adherence to CPAP and CBT-I.

Chapter 2 emphasised the importance of considering adherence not only as a multi-dimensional, but also as a biopsychosocial phenomenon (Aim2). As outlined in Table 23, Chapter 5 aimed to elucidate the differential physical and psychological effects on the relationship between CPAP use and symptomatic improvement. The patient-level meta-analysis of three randomised placebo-controlled crossover trials indicated that increased use of real CPAP conferred improvements in daytime sleepiness, but interestingly, so did increased use of placebo CPAP. As described in chapter 1, the relationship between adherence and outcome might be bidirectional, with increased adherence leading to better symptomatic improvements [the direction exclusively concentrated on in chapter 5], but improvements might also encourage increased machine use. Regardless of this point, the results suggest this (bi-directional) relationship is a result of both physiological and psychological effects [significant interaction effect]. Thus studies reviewed in chapter 1 exposing symptomatic improvement as a strong predictor of CPAP use, might stem from the actual physiological improvements in sleepiness, but also from a psychological expectation of benefit. The results also reveal that daytime sleepiness should not be purely considered a biomedical variable, but malleable to psychological effects or rather an expectation of benefit [placebo effect per se]. Thus the evidence for adopting a biopsychosocial approach to CPAP adherence emerges from two aspects of chapter 5 results: the significant interaction effect and placebo effect per se.

As outlined in chapter 2, the translation of a biopsychosocial approach of adherence into routine clinical practice requires the development and validation of questionnaires facilitating the identification of psychological and social variables (Aim 3). The final study in chapter 6 outlined this process for a scale assessing components of the transtheoretical model. Particular care was given to a number of issues to ensure this could be translated easily into clinical care. First of all, the scale had to be brief, ensure face validity and potentially transferrable to other sleep problems or health domains. To ensure brevity, there was a conscious decision against the use of multi-dimensional continuous measures of readiness to change, such as the 32-item version of the URICA.(512) Although the merit of this scale is the continuous measurement of readiness to change, however the brevity of an algorithm based scale with few items was deemed more appropriate for accomplishing the thesis aims.

Secondly, the scale had to warrant face validity and this was accomplished with the cognitive pretesting of the SOCSI (study 1). Future studies might want to test the face validity of the scale as rated by treating practitioners, however this was beyond the scope of the thesis. Although the SOSCI was developed and then successfully validated within the context of an insomnia population (study 2), certain steps were taken to ensure it could easily be adapted for the use in other sleep populations, or even other health domains. This included the consideration of open-box response formats, and the use of the term "action plan". This effort extends the aims of a BPS model of CPAP adherence to a holistic model of *adherence in general*. This though perhaps stems from the main strength of this thesis: the consideration of two sleep disorders, see below.

Before considering the strengths and limitations of the thesis in the next paragraph, it is worth briefly discussing the secondary aim of chapter 6: to provide information about measurement of adherence to SRT. With most individuals within 30 minutes of their threshold and rising time, and only small to moderate correlations with outcome, it is difficult to establish meaningful cut-offs for optimal adherence. Intuitively, 30 minutes seems a logical, and this is a figure used for cut-offs of insomnia symptoms (SOL, WASO). As described in the discussion of chapter 6., future dose-response studies might elucidate further on this matter.

7.2 Strengths/Limitations of the Thesis and Future Directions

Because the specific strengths and limitations of each study have been presented in the individual discussions of each chapter, only overreaching strengths and limitations of the thesis will be considered here, with remarks to possible translation into future studies/directions in this field.

The main strength of this thesis is the consideration of both OSA and insomnia. This has hopefully allowed for a more holistic approach to the phenomenon of adherence in general, by not becoming immersed in aspects that are only unique to one disorder. However, the two disorders have been considered as two separate conditions within this thesis. Yet, as highlighted in chapter 1, there is considerable overlap between these disorders. The introduction of CPAP might further exacerbate any underlying insomnia, or

contribute to a new incidence of insomnia and this might negatively impact on adherence to CPAP. Likewise, the use of CPAP might complicate adherence to components of CBT-I. Getting up in the middle of the night according to the quarter of an hour rule might seem even less appealing in full CPAP attire. Future studies need to take an integrative approach, not only to adherence, but also at the level of sleep disorders. Less "siloing" and dichotomising between physiologists and psychologists is vital, if the field of sleep medicine is to progress.(343, 365, 560) Related to this limitation is the aspect of conducting four very disparate experimental studies. This has both strengths and limitations in itself. There is a benefit at the level of the learning outcomes of the thesis, becoming skilled in various methodologies, including qualitative, patient-level meta-analysis and scale development, see Table 23. However simultaneously, this has made the translation of the experimental findings of each chapter into studies that follow on from and complement previous results slightly less likely. The results of all experimental chapters provide support for the adoption of a BPS approach; however conducting four disparate experimental studies also precludes the systematic evaluation of a BPS approach to either sleep disorder. Future studies should focus on the systematic exploration of possible interactions across all BPS domains, as outlined throughout this thesis.

In a concluding remark, this thesis has not exploited the uniqueness of sleep itself in understanding the phenomenon of adherence. Sleep is supposed to be restful, peaceful, and the introduction of something as intrusive as CPAP or sleep restriction and stimulus control is naturally going to meet with resistance.(see 561) Furthermore, the consequences of sleep deprivation/disturbances on mood in the long-term can lead to higher rates of depression and that is known to affect adherence to medication;(234) this might be a unique consideration for disorders of sleep.(562) Crucially, non-adherence to treatments for sleep disorders such as insomnia and OSA occur largely at a time where rational decision-making is impaired: at night. This aspect was briefly touched upon in chapter 3, however there is sufficient scope in future studies to understand how sleep might be present itself as a unique consideration for adherence. Furthermore, this has particular implications for interventions that incorporate non-rational aspects of human behaviour, such as habit-forming principles.(e.g., 437)

In reflection on this body of work, an exclusive focus on only one of these sleep disorders might have enabled a more comprehensive exploration of possible predictor variables, leading to a development of an interventions to improve adherence. The latter aspect has not been explored experimentally within this thesis. However, the definite advantages of the simultaneous consideration of both OSA and insomnia will hopefully allow for an interesting translation into further research, possible interventions, and clinical practice beyond this thesis.

7.3 Concluding remarks

In summary this thesis aimed to advance our understanding of contributing factors to the variance in adherence to treatments for OSA and insomnia. With the literature in both domains just at the cusp of agenda change, this thesis aimed to provide novel information about potential psychological and social variables, the interaction of variables from different domains of the biopsychosocial model of adherence and to develop tools to facilitate the assessment of these variables. Various avenues for future research to pursue have been suggested. Importantly, these need to consider adherence as a biopsychosocial phenomenon and begin to understand the unique consideration of the sleep state; after all,

> "Sleep that knits up the ravelled sleeve of care
>
> The death of each day's life, sore labour's bath
>
> Balm of hurt minds, great nature's second course,
>
> Chief nourisher in life's feast."
>
> -- William Shakespeare

Appendices

Appendix 1: Research Diagnostic criteria for Primary Insomnia Taken from Edinger et al.(92)

Research Diagnostic Criteria for Insomnia Disorder

A. The individual reports one or more of the following sleep related complaints:
   1. difficulty initiating sleep,
   2. difficulty maintaining sleep,
   3. waking up too early, or
4. sleep that is chronically nonrestorative or poor in quality.

B. The above sleep difficulty occurs despite adequate opportunity and circumstances for sleep.

C. At least one of the following forms of daytime impairment related to the nighttime sleep difficulty is reported by the individual:
   1. fatigue/malaise;
   2. attention, concentration, or memory impairment;
   3. social/vocational dysfunction or poor school performance;
   4. mood disturbance/irritability;
   5. daytime sleepiness;
   6. motivation/energy/initiative reduction;
   7. proneness for errors/accidents at work or while driving;
   8. tension headaches, and/or GI symptoms in response to sleep loss; and
   9. concerns or worries about sleep.

Research Diagnostic Criteria for Primary Insomnia

A. The individual meets the criteria for insomnia disorder.

B. The insomnia noted in A has been present for at least one month.

C. One of the following two conditions applies:
   1. There is no current or past mental or psychiatric disorder.
   2. There is a current or past mental or psychiatric disorder, but the temporal course of the insomnia shows some independence from the temporal course of the mental or psychiatric condition.

D. One of the following two conditions applies:
   1. There is no current or past sleep-disruptive medical condition.
   2. There is a current or past sleep-disruptive medical condition, but the temporal course of the insomnia shows some independence from the temporal course of the medical condition.

E. The insomnia cannot be attributed exclusively to another primary sleep disorder (e.g., sleep apnea, narcolepsy, or parasomnia) or to an unusual sleep/wake schedule or circadian rhythm disorder.

F. The insomnia cannot be attributed to a pattern of substance abuse or to use or withdrawal of psychoactive medications.

Appendix 2- Consent form for Qualitative Studies[1]

University of Glasgow | Sleep Centre

NHS Greater Glasgow and Clyde

Study Number:
Participant Identification Number for this study:

Consent Form

Title of the Project: Adherence in the Treatments of Disorders of Sleep and Wakefulness
Name of Researchers: Megan Crawford and Professor Colin Espie

1. I confirm that I have read and understand the information sheet and received my own copy dated the 12th of February 2009 for the above study. ☐
2. I have had the opportunity to consider the information provided, ask questions and have had these answered satisfactorily. ☐
3. I understand that my participation is voluntary and that I am free to withdraw at any time, without giving reason, without my medical care or legal rights being affected. ☐
4. I understand that the data collected during this study may be looked at by responsible individuals from the research team or from regulatory authorities where it is relevant to them taking part in research. I give permission for these individuals to have access to my records. ☐
5. I understand that the study will be tape recorded and audio data will be destroyed after transcription. ☐
6. I give permission to contact my GP and/or sleep specialist for information regarding this particular study. ☐
7. I consent to the results, which may be in form of direct quotes (anonymised form), of this study being published in relevant journals ☐
8. I would be interested in being contacted in the furture from the University of Glasogow Sleep Centre regarding future studies. ☐
9. I agree to take part in the above study. ☐

Name of Participant	Date	Signature
Megan Crawford	20th of April 2009	
Researcher	Date	Signature

Version 2: 12/02/2009

[1] Consent forms for insomnia and OSA populations were identical. Layout differed according to research setting (e.g. Glasgow University and NHS headings were replaced with Royal Prince Alfred Hospital and Woolcock Institute logos, see appendix xxx).

Appendix 3- Participant Information Sheet for Qualitative Study- OSA

ROYAL PRINCE ALFRED HOSPITAL
A tradition in excellence since 1882

WOOLCOCK INSTITUTE of MEDICAL RESEARCH

Adherence in the Treatments of Disorders of Sleep and Wakefulness

Participant Information Sheet for Obstructive Sleep Apnoea

Purpose of the Study

Obstructive Sleep Apnea (OSA) is a common sleep disorder which has major negative health implications and is a serious risk factor for cardiovascular disease. Continuous Positive Airway Pressure (CPAP) is the most effective and widely used treatment choice as it reduces the number of nocturnal respiratory events or the number of times you stop breathing during sleep, improves the quality of sleep and reduced daytime sleepiness. However many individuals do not take up their CPAP treatment for a number of reasons.

Previous testing (an overnight sleep study) has shown that you have sleep apnea and due to the severity of your disease CPAP was recommended as the first choice of treatment following screening and discussion with your sleep physician.

You are invited to participate in a research study aiming to understand the experience implementing the recommendations given by your practitioners about using CPAP. Understanding the factors that enable participants to start and maintain treatment recommendations is important in helping us further improve treatment interventions.

Voluntary Participation and Withdrawal from the Study

You have a right not to participate in the study and you can withdraw at any time. A decision not to participate or to withdraw from the study will not affect your current or future treatment or your relationship with the University of Sydney or Royal Prince Alfred Hospital or any of the participating hospitals or institutions co-operating in this study or any person treating you.

Description of Study and Risks

If you decide to participate in this study, you will be invited to come to the Woolcock Institute of Medical Research, where firstly you will be asked to sign the Participant Consent Form. Thereafter you will, as part of routine treatment, undergo a CPAP titration study, which is similar to your overnight diagnostic sleep study. This time however you will be using a CPAP machine to determine what water pressure is required to stop your 'stopping breathing' episodes. Treatment as Usual (TAU) also consists of a standard education presentation of 12 slides, which describe normal sleep; sleep staging; what is obstructive sleep apnoea and how CPAP works (this education session is given before your titration study). In addition to this routine treatment procedure you will be asked to take pat in one-on-one interviews with the research co-ordinator at 3 different time points. The first interview will be on the same day you come in for your CPAP titration (so you will need to come in a few hours before your titration study to meet for the interview). You will be then, as for all clinic patients, asked to purchase and pick up your own CPAP machine, mask and tubing after you have received a script from the sleep unit staff stating the information you will need. After you pick up your mask, you will be asked to contact the research investigator, and you will then be invited to come for 2 additional interviews, the second interview will be held 1 week and the third interview 3 months after you pick up your mask.

The interviews will last approximately 1 hour and will be one-on-one semi-structured interviews. It will resemble mostly an informal conversation; however some questions regarding your experience with your OSA and CPAP treatment will be prepared. The interview will be tape-recorded and the data will be deleted after transcription. The results of this study will be published in a relevant journal so that the general public is also aware of these findings. These reports/publications may include quotes from the participants, however all information regarding your/their identity will not be revealed in these. Also,

anything you say will not affect your subsequent treatment delivery. After each interview you will also be asked to fill in some questionnaires (described below). All the information that is collected, during the course of the study, will remain strictly confidential. This means that all information will be kept secured in locked filling cabinets and only the researcher will be able to access these, you will be given a unique code that will ensure confidentiality and anonymity of all your data. The researchers would like to have access to your medical record to obtain information relevant to this study.

Description of Questionnaires[2]

- Epworth Sleepiness Scale (ESS) presents 8 everyday situations and you are asked to rate your chance of dozing in these circumstances.
- Hospital Anxiety and Depression Scale (HADS) is a validated 14-point scale that asks you to rate your mood and feelings in relation to a number of different statements.
- Pittsburgh Sleep Quality Index (PSQI) is a validated 11 point questionnaire widely used in sleep research which assesses sleep quality, insomnia symptoms, medication use and other sleep disorders.
- The Illness Perception Questionnaire (IPQ-R) asks you about your perceptions about your illness in terms of consequences, time-line, cure/control, coherence and emotional representation.
- Transtheoretical Model Scale (TM) is a validated questionnaire and assesses variables involved in changing behaviour.
- The Daytime Functioning Scale is a 12 item scale that ask you to rate the importance of various effects of your sleep problem on your daytime functioning .
- The Glasgow Sleep Impact Inventory (GSII) is a rating scale of the impact your sleep problem has on your life.
- The University of Rhode Island Change Assessment Scale-Insomnia (URICA) is a scale adapted from the validated URICA used to assess individuals' readiness to change.
- Functional Outcomes Sleep Questionnaire (FOSQ) is a quality of life scale developed specifically to assess lifestyle difficulties associated with any sleep disorders you may or may not experience.
- The Social Cognitive Theory scale (SCT) is a validated questionnaire with items asking about the outcome expectancies you have, ratings of your self-efficacy and the social support you have.

While we intend that this research study furthers medical knowledge and may improve treatment of OSA in the future, it may not be of direct benefit to you.

Confidentiality and Disclosures of Information

[2] These questionnaires were part of secondary analysis are not included in this thesis.

Any information that is obtained in connection with this study and can be identified with you will remain confidential and will be disclosed only with your permission or except as required by law. If you give us your permission by signing the Participant Consent Form, we plan to publish the results in medical or scientific journals and we may also reanalyse the data at some later stage. In any publication, information will be provided in such a way that you cannot be identified.

If you have any questions, please feel free to ask us. If you have any additional questions concerning the availability of medical care, or if you think you have experienced a research related illness, injury or emergency, please immediately contact the Principal Investigator, Professor Ron Grunstein, on (02) 9114 0007, the co-investigator Dr Delwyn Bartlett, on (02) 9114 0460, or the research coordinator/officer for this project, Megan Crawford (02) 9114 0456.

Ethics Approval

This study has been approved by the Ethics Review Committee (RPAH) of the Sydney South West Area Health Service. Any person with concerns or complaints about the conduct of a research study can contact the Secretary on (02) 9515 6766 and quote protocol number xxx.

This information sheet is for you to keep.

Appendix 4- Interview schedule for Qualitative Studies

Preamble to Semi-Structured Interview

Knowing about people's experiences of a sleep problem and their treatments can be very useful in helping us to improve treatment experience. Often research uses questionnaires and designs experiments to find out how individuals react to treatments and how and why they try to follow treatment recommendations. Another research tool is to ask the individual to report of their experience of the sleep problem and the treatments. This provides unique information that questionnaires and experiments do not always do. Therefore I would like to ask you a few questions about your sleep problem, how you experience(d) treatment, and how you tried to follow the advice given by your doctor/therapist. I will start with some general questions and then follow with some more specific ones. All the information you give will remain anonymous and you are free to withdraw from the study/stop the interview at any time. The interview will be audio-taped, but you may disagree with this, if you wish for it not to be recorded.

The Disorder and Treatment

1. Can you describe the history of your sleep problem, from when it started to the beginning of treatment?

2. How did you feel about having sleep apnoea/insomnia?
   PROMPT: What did it mean to you?
   PROMPT: What kind of implications did it have for you?

3. What did you feel at first when this treatment was suggested?
   PROMPT: What were your thoughts/knowledge about the treatment before it was suggested?
   PROMPT: What did you expect from the treatment?

4. What did you feel when you first tried it out?
   PROMPT: physically, emotionally, mentally.

5. What is using CPAP/CBT like for you now?
   PROMPT: physically, emotionally, mentally.
   PROMPT: How are things different for you now?

6. How does CPAP/CBT affect your every day life?
   PROMPT: Is it positive/helpful or more a burden?

7. How do you feel about your sleep problem now?
   PROMPT: What does it mean to you?
   PROMPT: What kind of implications does it have for you?

Implementing Treatment

8. Can you describe a typical night when using your CPAP machine/CBT?
   PROMPT: What do you think influenced your behaviour?

9. Can you describe a typical night when you did not use your machine/CBT?
   PROMPT: What do you think influenced your behaviour?

10. Different people behave differently when asked to follow a set routine? Do you have any thoughts on that?
    PROMPT: Can you give an example of your own experience?
    PROMPT: Is this similar to your experience with CPAP/CBT?

11. If you could choose one thing to change that would help you follow a set routine to the requirements, what would that be?
    PROMPT: Can you give any further examples?

12. What do you think your medical practitioner emphasises as important in following treatment recommendations?

13. Is there anything you would like to add to the information given so far?

Appendix 5: Poster preseted at the American Professional Sleep Societies Conference, Minneapolis, 2011

Are Treatment Acceptability and Health Value Associated with Adherence to Treatments for Obstructive Sleep Apnea

Megan R. Crawford[1,2,3] Greenwood, A.[2] Bartlett D.J.[2,3] Espie, C.A.[1,3] Grunstein, R.R.[2,3] Cistulli, P.A.[2,3]

1 University of Glasgow Sleep Centre, Scotland, UK, 2 Woolcock Institute of Medical Research, University of Sydney, Australia
3 NHMRC Centre for Integrated Research and Understanding of Sleep (CIRUS)

Contact: m.crawford.1@research.gla.ac.uk

Background

- Mandibular advancement devices (MAD) offer an alternative to Continuous Positive Airway Pressure (CPAP) for Obstructive Sleep Apnea (OSA).
- Some evidence is available regarding the disease phenotype (e.g. AHI, upper airway physiology) that might predict which individuals respond best to either treatment.
- It is unclear which psychological variables might predict adherence.
- Treatment effectiveness might be a predictor of adherence to treatment, but self-efficacy could mediate this relationship, especially for CPAP- a cumbersome, demanding treatment.

Aims

- To establish a psychological profile that might predict adherence to treatments for OSA.

Methods

Measures and Subjects

- Thirty-four individuals with OSA were conveniently sampled from a large randomised controlled cross-over study (n=108) evaluating the effectiveness of MAD versus CPAP.
- Treatment Acceptability for both MAD and CPAP, Health Value (all variables split into high vs. low based on median) and general self efficacy were measured. We examined the effects of these variables on adherence to both treatments.

Table 1: Characteristics of the sampled 34 patients

Number of Participants	34
Age (Years)	47 (range25-66)
Gender (female)	4 (11%) female
Baseline Epworth Sleepiness Scale	8 (SD 4)
Baseline Apnea Hypopnea Index	22 (SD 11)
Prescribed CPAP pressure (cmH_20)	10 (SD2)
Maximal Protrusion with MAD (%)	152 (SD 67)

Statistical analysis

- Differences in Treatment Acceptability (each subscales: acceptability, convenience, severity of side effects, effectiveness) was examined for CPAP and MAD using a student t-test or the equivalent non-parametric test.
- Analysis of Co-Variance (ANCOVA) was computed to establish the effect of treatment acceptability and health value on adherence to CPAP and MAD, controlling for self-efficacy.

Results

Differences in treatment acceptability between CPAP and MAD

- **Post-Acclimatisation Period**
  - There was no difference between the acceptability subscales, except for Convenience. MAD (mean=2.5, SD=0.9) was considered more convenient than CPAP (mean=1.9 SD=0.9), t(32)= -2.6; p<0.01, (Figure 1). These effects were retained post treatment (Figure 2).

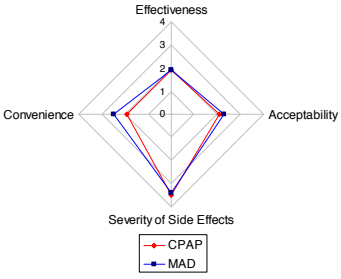


Figure 1: POST ACCLIMITISATION
Differences between Treatment Acceptability Subscales for MAD and CPAP

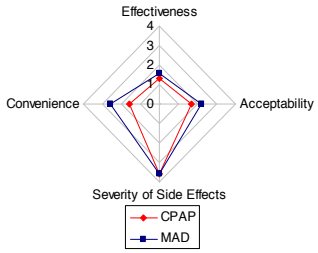


Figure 2: POST TREATMENT
Differences between Treatment Acceptability Subscales for MAD and CPAP

Effects of Acceptability and Health Value on CPAP adherence

- There was a trend for an effect of treatment acceptability on adherence levels in the CPAP phase;
  - Mean adherence for the high scoring acceptability group (mean = 4.6, SD = 2.5) was higher than for the low scoring acceptability group (m=3.4, SD=2.0), t (30) =-1.7, p=0.08, representing a medium effect, d=0.6 (Figure 3).
  - This effect remained after controlling for self-efficacy.
- Health value had a large effect (d=0.8) on adherence in the CPAP phase; higher mean adherence was evident in those that viewed their health as important (m=4.8, SD=2.1) as compared to those who viewed it as less important (m=2.9, SD=2.5), t (31) =-2.4, p<0.05 (Figure 3).
- Interestingly the relationship between adherence and psychological variables was not observed in the MAD phase.

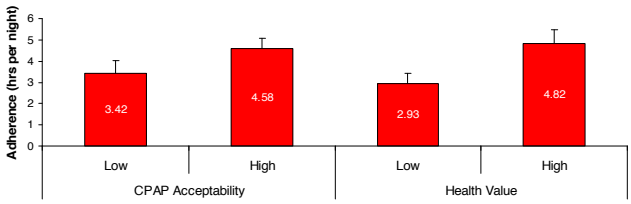


Figure 3: Effects of Treatment Acceptability and Health Value on adherence to CPAP. Bars represent mean value of adherence with standard error bars.

Conclusions

- This study reveals that acceptability of CPAP might have an effect on treatment use, yet contrary to our hypothesis, this relationship was not mediated by the belief in one's own capabilities.
- Individuals who value their health are increased CPAP users.
- Interestingly, these relationships are not reflected in MAD use, potentially, because this treatment is seen as more convenient than CPAP, and thus psychological variables have less of an impact on adherence.

Acknowledgements

Funding: Australian and UK MRC

Appendix 6- Participant Information Sheet for Qualitative Study- Insomnia

University of Glasgow | Sleep Centre

Version 2: 12/02/2009

Participant Information Sheet- Insomnia

Title: Adherence in the Treatments of Disorders of Sleep and Wakefulness

You are being invited to take part in a research study. Before you decide whether or not to take part, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and discuss it with other if you wish. Ask us if there is anything that is not clear or if you would like more information. Take time to decide whether or not you wish to take part.

The Study

It is a research study being conducted at the University of Glasgow Sleep Centre . This study is designed to understand the experience of having a sleep problem, what treatments for your sleep disturbances are like and your experience in trying to follow the recommendations of your doctor. In order to understand this kind of information, researchers carry out interviews to ask people about their experiences. This is also the nature of this study; therefore, you will be invited to the University of Glasgow Sleep Centre for an interview, where the researcher will ask about your experiences with your sleep problem and the treatment. This will hopefully give us more information of how to improve treatments and how to help people trying to follow treatment advice.

Do I have to take part?

Taking part in the research is entirely voluntary. This means that it is up to you to decide whether or not you would like to take part. If you do not wish to take part it will not affect the case or your rights in any way. If you decide to take part in the study, you can keep this information sheet and return the consent form and your responses to the questionnaires enclosed. If you do decide to take part, you can change your mind and withdraw from the study at any time, even after the study has finished.

What does taking part involve?

If you decide to take part, you will be invited to come to the University of Glasgow Sleep Centre at the Southern General for an interview. You will meet with the researcher who is interested in finding out about your experiences with your sleep problem and the treatments. A few questions will be prepared that the interviewer will ask, but generally the interview will be more like an informal conversation. The interview will last approximately one hour. Afterwards you have the opportunity to answer a few questionnaires, which will take a few minutes of your time and we will also ask you for some feedback on the interview that might improve future interviews.

Would my results be kept confidential? What will happen to the results of the study?

All the information that is collected, during the course of the study, will remain strictly confidential. This means that all information will be kept secured in locked filling cabinets and only the researcher will be able to access these, you will be given a unique code that will ensure confidentiality and anonymity of all your data. The interview will be tape-recorded and the data will be deleted after transcription. The results of this study will be published in a relevant journal so that the general public is also aware of these findings. These reports/publications may include quotes from the participants, however all information regarding your/their identity will not be revealed in these. Also, anything you say will not affect your subsequent treatment delivery.

What are the potential benefits of taking part?

Taking part will hopefully help us broaden our knowledge of how patients experience their sleep problem and treatments. This in turn will provide us with information of how to improve treatments and how best to support the patients during the course of treatment.

If I decide to take part what happens next?

Apart from this Participant Information Sheet you will also find a consent form, which you will need to return and send back to the Sleep Centre in the self-addressed envelope. Enclosed is also a leaflet with Frequently Asked Questions to help you with any problems that may arise.

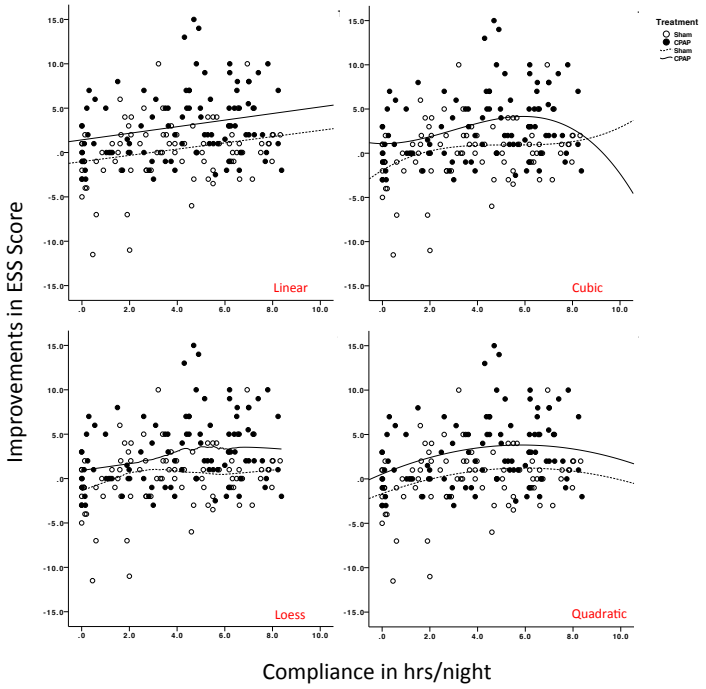
Thank you for reading this information. If there is anything you are not clear about or if you have any questions regarding this study you can e-mail the researcher at

m.crawford.1@research.gla.ac.uk or call:

University of Glasgow Sleep Centre Recruitment Number: 07503730769

If you would like some independent advice from someone who is not involved in the study, please contact Dr Maria Gardani +44 (0)141 232 7700 or M.Gardani@clinmed.gla.ac.uk

Appendix 7: Supplementary Figure: Dose-response curves for CPAP use and ESS improvement



Appendix 8- Description of the Transtheoretical Model

Stage of Change

In the early versions of their model Prochaska and DiClemente described how smokers pass through distinct stages when considering and implementing change(504, 563). In further refinements of their model through experimental studies, they identified 5 stages of change: pre-contemplation, contemplation, preparation, action and motivation.(564-567)

Each stage represents an amalgamation of the indvidual's current behavioural state and intention to change. In precontemplation stage, the unhealthy behaviour (e.g. smoking) is the status quo and the individual has no intention of changing, at least not in the near future (usually 6 months). At the next stage, the individual reaches a point of ambivalence; the importance for the new behavioural pattern starts to emerge and the individual begins to contemplate behaviour change. It is a period of extreme instability and often individuals become stuck in this stage for a lengthy period of time.(504) At the point at which the benefits for the new behaviour outweigh the costs, the individual begins to make concrete plans for change within the near future. Irregular implementation of behaviour in the past year is often described as characteristic of this preparation stage(503). The consistent behavioural implementation occurs in the action stage, and once the new behaviour becomes the status quo for a period of time (often described as 6 months), the maintenance stage is reached. These temporal distinctions are somewhat arbitrary, but necessary to place individuals into distinct categories.(556)

Although the theoretical progression through these stages reflects a linear increase in readiness to change, the natural progression through these stages is described as cyclical, often portrayed as the revolving door principle (after moving through to later stages, relapsing back to earlier stages prior to continued progression).(504, 564) Interestingly, their experimental work

suggests that individuals learn from their experience with only 15% of smokers relapsing to the earliest stage of pre-contemplation(503), see also Figure 16.

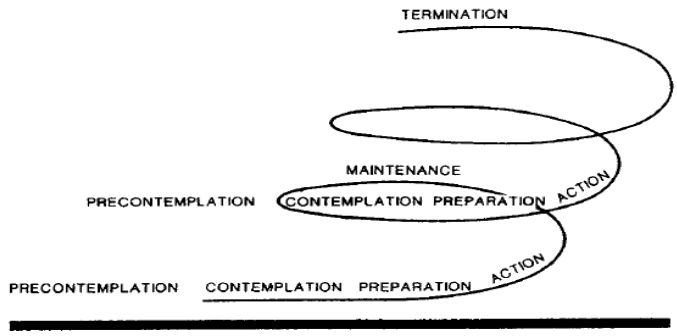


Figure 16: A spiral model of the stages of change.
Source: Prochaska et al. 1992(564)

Process of Change

The model further specifies distinct processes that initiate the transition to theoretically superior stages.(502) Modelled again on existing theories, they describe 10 internal and external strategies that can be implemented to facilitate stage transition. These are classified as overt (behaviour) or covert (cognitive) and it is claimed that the covert processes are involved in the transition through the earlier stages to initiate contemplation of change, whereas overt processes are involved in facilitating maintenance of change.(547) See Table 24.

Table 24: Processes of Change

	Process	Description
Covert (Cognitive)	Consciousness raising	Making use of informational resources to raise awareness of the new behavioural pattern
	Self-reevaluation	Evaluation of the new behavioural pattern against personal values
	Environmental reevaluation	Evaluation of the impact on environmental context (e.g. family)
	Emotional arousal/dramatic relief	Considering the emotional impact of the potential behavioural change
	Social liberation	Becoming aware of the social norms surrounding the old and new behavioural pattern
Overt (Behavioural)	Self-liberation	Making firm commitments for change, take responsibility and strengthen belief in own capabilities for change
	Conditioning/counter conditioning	Pursue situations that facilitate behaviour change
	Stimulus generalisation/control	Removal of stimuli that elicit old behavioural patterns
	Reinforcement management	Seeking rewards for successful behavioural implementation
	Helping relationships	Utilise social support to facilitate the implementation of the new behaviour

Markers of change: Decisional Balance and Self-efficacy/Temptation

With the precision of their model, Prochaska and colleagues included markers of change: the decisional balance and self-efficacy/temptation. The decisional balance was modified from Janis and Mann's decision making theory(568) and describes change as a process of weighing up potential benefits against costs of the new behavioural pattern. Visually plotting the importance pros and cons for behaviour change across the stages in Figure 17 highlights how the costs of the new behaviour outweigh the benefits in the preparation stage, an equal balance in the contemplation stage; and finally a cross-over occurs, when the pros for change outweigh the cons for change in the remaining stages. (569)

Lastly, the TTM also gives prominence to self-efficacy; a concept originally framed by Albert Bandura, which has found its way into numerous theoretical models and describes the individual's self-reflected efficacy expectations. That is, the belief in one's capabilities to achieve set goals, especially when confronted with certain barriers(339, 570). The TTM states that with progression through the stages, the impact of self-efficacy increases in a linear fashion.(496) A large evidence base supports the significance of this concept in predicting maintenance of change.(571, 572). This is not to say that self-efficacy is not also imperative in the earlier stages -a deflated belief in one's abilities might inhibit contemplation and initiation of change(573) - however there is a complex interaction between self-efficacy, stage of change and processes of change.(496) Self-efficacy is a marker of increased use of cognitive change processes in the earlier stages. However, in the action and maintenance stages, increased self-efficacy is associated with less use (or need to use) of behavioural change processes;(573) hence, self-efficacy can be described as both a marker, mediator and moderator of change dependant on stage.(574)

Temptation is described as the counterpart to self-efficacy and is found to correlate negatively with self-efficacy, however is not its exact mirror image: the temptation to engage in the old behavioural pattern can be high, despite the individuals increased confidence to resist that temptation.(496)

Figure 17 is a visual representation of the processes and markers of change and how these relate to the individual stages.

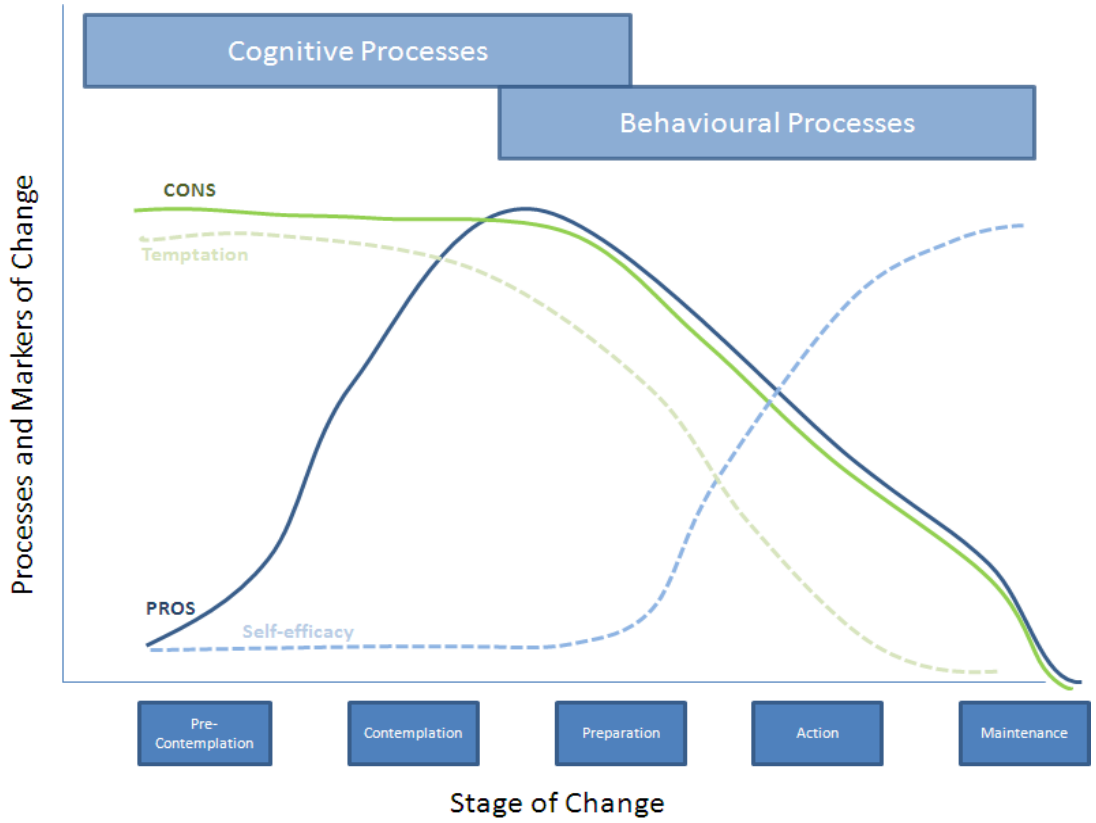


Figure 17: TTM overview
Relationships between processes and markers of change. With progression through the stages, pros outweigh the cons for change (the influence of the pros on change diminishes once behaviour is implemented and maintained. The TTM model states that the importance of self-efficacy increases with stage progression, whereas temptation is reduced. Cognitive processes induce change in earlier stages, wheras behavioural processes are most important in transition between the latter stages. Adapted from Velicer et al. 1998.(575)

Measuring stage of change

In experimental studies individuals have been assigned to stages using algorithms or measures with stage-specific subscales. Staging algorithms present the individuals with a series of questions and responses classify individuals into one stage only. An alternative is to present a statement for each stage and individuals are asked to indicate which they agree with most. These statements are often modelled on published stage descriptions in smokers(566, 567) see Table 25.

Table 25: Description of stages used in staging algorithms

Pre-contemplation	'I don't currently have a problem, and I am not willing to change within the next 6 months'
Contemplation	'I have a problem, I am thinking of changing within the next 6 months'
Preparation	'I am planning to change within the next 30 days'
Action	'I have implemented the new behaviour within the last 6 months'
Maintenance	'I have implemented the new behaviour for more than 6 months'

This method is particularly useful, because these algorithms are short and easily integrated into both cumbersome research protocols and clinical settings. There is emerging evidence for the use of this approach in domains other than smoking cessation, for example drug use(576) or medication adherence.(523, 549)

Three main multidimensional measures have been used, whereby stage allocation is determined either by highest scoring subscale or cluster analysis. The University of Rhode Island Change Assessment(URICA, 512) the Readiness to Change Questionnaire(RCQ, 577) and the Stages of Change Readiness and Treatment Eagerness Scale,(SOCRATES, 578) are measures that have been frequently used in research administering TTM dimensions. The advantage of these scales over the algorithms is the possibility of generating both a continuous and categorical outcome variable. However, the key criticism of the use of these measures is the lack of overlap with the 5 stages of the transtheoretical model, with few replicating the cluster profile described in the model.(579)

Appendix 9: Interview Schedule- Excerpt

BOLD text indicates instructions for the interviewer

Italicised text indicates instructions to be read to the participant

Underlined text indicates the material being tested.

Cognitive Interview:

Date ___/ ___/ ____ Interview # _____ Interviewer Initials ___ ___ ___

Enter start time of interview __ __ : __ __

Read these instructions in their entirety or paraphrase them but include elements 1 to 7

Thanks for coming in. Let me tell you a little more about what we'll be doing today.

1. *We're testing a new questionnaire with the help of people such as yourself.*
2. *I'll ask you questions and you answer them, just like a regular survey.*
3. *However, our goal here is to get a better idea of how the questions are working. So I'd like you to think aloud as you answer the questions- just tell me everything you are thinking about as you go about answering them.*
4. *At times I'll also stop and ask you more questions about the terms or phrases in the questions and what you think a questions is asking about. I'll also take notes.*
5. *Please keep in mind that I really want to hear all of your opinions and reactions. Don't hesitate to speak up whenever something seems unclear, is hard to answer, or doesn't seem to apply to you.*
6. *Finally we'll do this for an hour unless I run out of things to ask you before then.*
7. *Do you have any questions before we start?*

Practice task

I'd like to begin with a practice questionnaire so you get a feel for what is required in the real interview. First of all, I would like you to answer the questions on this sheet. After you have answered them, please mark your comments on the questionnaire with a pen. Mark down any difficult words or any phrases which are unclear. Please keep in mind that I

want to hear all of your opinions, so don't hesitate to make as many comments as you want.

Give the questionnaire to the participant and let them fill it out on their own.

Practice question 1: How many windows are there in the house or apartment where you live?

Practice question 2: How difficult was it for you to get here to do the interview today: very difficult, somewhat difficult, a little difficult, or not at all difficult?

Now go through question and answer each individually.

Okay now I will ask you a series of questions about the questionnaire you just filled out. Remember to try to think aloud as you answer.

So starting with question 1, What is this question asking you?

[Probe as necessary]: I see you answered [answer]. How did you come up with that answer?

Moving on to question 2, What is this question about?

Looking at the answer options, what do the terms "somewhat difficult" and "a little difficult mean" to you? Are they different?

[Probe as necessary]: Tell me about your answer, why did you say [answer]?

Make a note of any difficulties the patient has with this task

Have they grasped thinking aloud?

Are they reticent about making critical comments?

Any Other issues?

Appendix 10: Conditioned Probes

The following conditioned probes were used in the cognitive pretesting if necessary:

Conditioned Probing:

Condition	Conditional Probe
1. Subject cannot answer or does not know the answer	"What was going through your mind as you tried to answer the question?"
2. Subject answers after a period of silence.	"You took a little while to answer that question. What were you thinking about?"
3. Subject answers with uncertainty using explicit cues such as "um," "ah" chaning an answer, etc.	"You seem to be somewhat uncertain. If so, can you tell me why?" "What cause you to change your answer?"
4. Answer is contingent on certain conditions being me, e.g. "I'd say about 25 times if you don't need a super precise answer."	"You seem a little unsure. If so, can you tell me why?"
5. Erroneous answer; verbal report implies misconception or inappropriate response process.	Clarify respondent's understanding of the particular team or the process used. For example, if the respondent appeared to misunderstand the word "manage", probe the term ("so you don't manage any staff")
6. Subject requests information instead of providing an answer.	"If I weren't available or able to answer, what would you decide it means?" "Are there different things you think it might mean? What sorts of things?"

Taken from(Conrad & Blair (2001, cited in 525)

Appendix 11: SOCSI Final Version

SOCSI-TREATMENT VERSION 2

People often find it difficult to stick to action plans consistently, even after they have agreed their therapy goals. This sheet is a step-by-step way of recording what your goals are at the moment; and how you feel about sticking to the action plan between now and your next treatment session. Please complete the 6 steps below.

ID: Date: Session 1

STEP 1. What action plan (e.g. relaxation, sleep schedule) did you discuss with your therapist, in order to help with your sleep problem?

STEP 2. Please follow through the next set of questions.

Q.1. Do you currently consistently stick to the action plan?
a. Yes (Go to Q2)
b. No (Go to Q3)

Q.2. Have you been consistently sticking to the action plan…

i. within the last 4 weeks
ii. and for longer than 4 weeks

Office Use

A
M

Q.3. Are you thinking about consistently sticking to the action plan?
a. Yes, I will start doing so from now on
b. Yes, I am considering doing so in the future
c. No, I am not considering doing so in the future

Prep
C
PreC

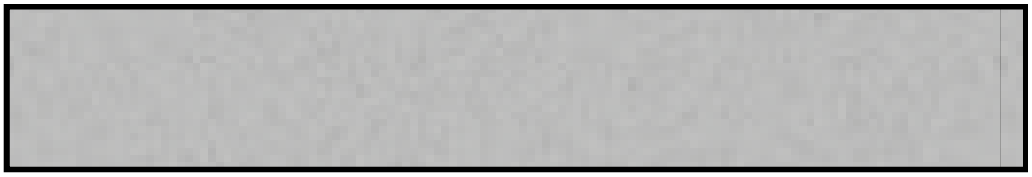
PLEASE TURN THE PAGE

STEP 3. How confident are you about sticking to the action plan?

Not at all confident	Slightly confident	Moderately confident	Very confident	Totally confident
0	1	2	3	4

STEP 4. What do you see as the advantages of sticking to the action plan?

STEP 5. What do you see as the disadvantages of sticking to the action plan?



STEP 6. What can you do to help you stick to the action plan?

Thank you very much for completing this survey!

Appendix 12: Initial Screening Form

PLEASE MAKE SURE THAT ALL OF THE FORM IS FILLED IN CLEARLY INCLUDING WHO TOOK THE CALL AND THE DATE AND TIME. NOTES SHOULD BE KEPT ON SEPARATE PIECE OF PAPER.

Source

How did you find out about the University of Glasgow Sleep Centre?	
Why have you contacted us?	
Method of initial contact (mobile, email, office phone)?	

Personal

Full Name:	*Date of Birth:*	*Age:*
Telephone:	*Address:*	
Alternative Telephone:		
When is a good time to call?		
What GP practice do you attend, and who is the GP you normally see?		

Sleep

Do you have difficulty sleeping at the moment? (Y/N)	
Are you generally satisfied with the amount of sleep you get (Y/N)	
Have you always been a poor sleeper? (Y/N)	
How long have you had a sleep problem?(yr)	
Do you have difficulty falling asleep? (Y/N)	

How many nights per week do you have difficulty falling asleep? (out of 7)	
How long does it normally take you to fall asleep?(min)	
Do you experience sleep disturbance because of waking up during the night?(Y/N)	
How many nights per week do you have a difficulty with waking up during the night?(out of 7)	
How long are you normally awake during the night, in total? (min)	
What time do you normally go to bed? (time)	
What time do you normally get up?(time)	
How long do you normally sleep?(hr/min)	
Do you have any other difficulties with your sleep (e.g. restless legs, breathing problems, sleep walking)?	
Do you work shifts, night shifts?	
Roughly, how many units of alcohol do you drink per week? (Remember: One standard (175ml) glass of wine = 2 unit One pint of standard lager = 2.3 units Spirit & Mixer = 1 unit)	
Does your sleep disturbance affect how you feel and function during the day (e.g. fatigue, sleepiness, concentration, memory, mood, motivation, irritable, work/social functioning etc.). If yes, specify most salient.	

Health

Do you keep in good health physically? (Y/N)	

What physical health problems do you have (if applicable)?
What medicines do you take for your physical health? (if applicable)
Do you keep in good health mentally? (Y/N)
What mental health problems do you have (if applicable)?
What medicines do you take for your mental health? (if applicable)
Do you give your consent for us to contact your GP if necessary regarding your health?

Suggested Algorithm to Screen for Sleep Disorder Other Than Insomnia.

Ask the lead question, and then proceed with supplementary only if answer is 'yes'.

1. Narcolepsy ***a. Do you sometimes fall asleep in the daytime completely without warning? (YES/NO)*** *b. Is it literally impossible to resist 'sleep attacks' during the day?* *c. Do you have collapses or extreme muscle weakness triggered by extreme emotion?* *d. Do you have visual hallucinations, either just as you fall asleep or when you wake in the morning?* *e. Are you paralysed and unable to move when you wake up from your sleep?* *[Possible narcolepsy: 1a¼''TRUE'' AND (1b OR 1c OR 1d OR 1e¼''TRUE'')]*
2. Sleep breathing disorder ***a. Are you a very heavy snorer? (YES/NO)*** *b. Does your partner say that you sometimes stop breathing?* *c. Do you often wake up gasping for a breath?* *d. Are you often excessively sleepy during the day or fall asleep without wanting to?* *[Possible sleep breathing disorder: 2a¼''TRUE'' AND (2b OR 2c OR*

2d¼''TRUE'')]

3. PLMS/ RLS
a. Do your legs often twitch or jerk or can't keep still in bed?(YES/NO)

b. Is it very difficult to get to sleep because of repeated muscle jerks?
c. Do you frequently wake from sleep with sudden jerky movements or with a compulsion to move your legs?
d. Do you simply have to get out of bed and pace around to get rid of these feelings?
[Possible PLMS/ RLS: 3a¼''TRUE'' AND (3b OR 3c OR 3d¼''TRUE'')]

4. Circadian Rhythm Sleep Disorder
a. Do you tend to sleep well but just at the ''wrong times''? (YES/NO)

b. Can you sleep well enough, but only if you stay up very late?
c. Are you in a very sound sleep at normal waking time and could sleep on for hours more?
d. Can you sleep well enough, but only if you go to bed very early?
e. Do you wake very early, bright and alert and no longer sleepy?
[Possible CRSD: 4a¼''TRUE'' AND EITHER (4b AND 4c¼''TRUE'') OR (4d AND 4e¼''TRUE'')]

5. Parasomnia
a. Do you have unusual behaviours, like sleepwalking, associated with your sleep that trouble you or that are dangerous?(YES/NO)

b. Do you sleepwalk frequently and run the risk of injuring yourself or others?
c. Do you have frequent night terrors when you are extremely distressed but not properly awake?
d. Do you act out your dreams and risk injuring yourself or others?
e. Do you have terrible recurring nightmares?
[Possible parasomnia: 5a¼''TRUE'' AND EITHER (5b OR 5c OR 5d OR 5e¼''TRUE''

If YES to b. or c.
Ask : Do you currently experience these?(YES/NO)
: How often in the past 6 months?

Appendix 13: Sleep/Bed Restriction Protocol

University of Glasgow | Sleep Centre

Bed Restriction Therapy

Information Manual

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Introduction

This information manual aims to help describe, in detail, bed restriction therapy, particularly the theory behind how it works, how to implement it practically, and any frequently asked questions you may have surrounding its use. If, after reading this information, you would still like to know more about Bed Restriction Therapy for insomnia please ask one of the researchers at the University of Glasgow Sleep Centre.

Regulation of sleep

Sleep is an automatic process and therefore out of our own direct, voluntary control.

Whether awake or asleep we are at the mercy of two biological processes:

(1) Sleep Homeostasis, commonly known as 'Sleep Pressure'

(2) The Circadian Rhythm, otherwise known as the 'Body Clock'

These two processes work in harmony to promote good consolidated sleep at night, and it is the goal of Bed Restriction Therapy to help restore the functioning of these two processes to enable undisturbed, good quality sleep to occur.

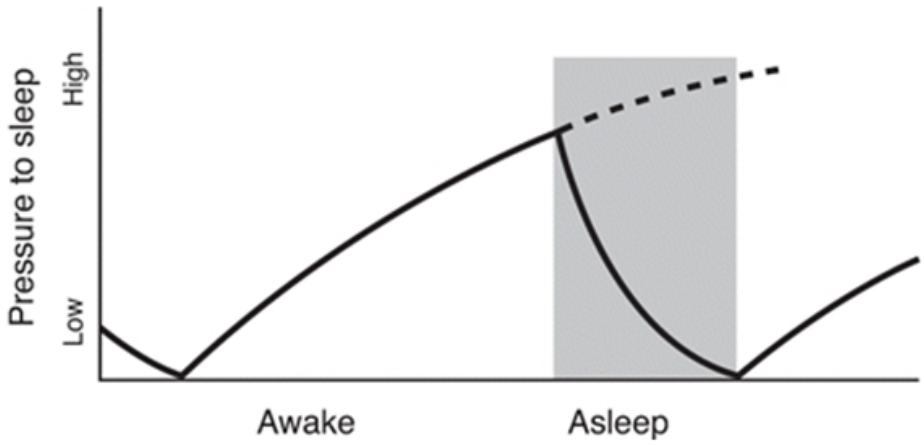
Below we describe how sleep pressure and our body clock work across a normal 24-hour day. By understanding how these processes we are able to find out what alterations may occur in insomnia, and crucially how these can be restored and improved with bed restriction therapy.

Sleep Pressure

Sleep pressure can be thought of as the brains pressure and need for sleep, which becomes greater with the increasing amount of time that we are awake. In this way, the pressure to sleep is directly related to the amount of time that we have been awake. For example, when we wake-up in the morning after a good night's sleep, we will have a very low sleep pressure or 'need to sleep'. As we continue throughout the day, sleep pressure will begin to accumulate (a bit like an hourglass egg-timer). Look at the diagram below which illustrates this increasing sleep pressure over the waking day. At the end of a full day, at bedtime, we will have a great amount of pressure to sleep. By going to bed and having another good night's sleep, then sleep pressure will be reset for the start of the next day.

Diagram of Sleep Homeostasis (Sleep Pressure)

The diagram below shows sleep pressure over the course of a day. The dashed line (---) shows that by staying awake later than your normal bedtime, you will continue to accumulate sleep pressure.



Circadian Rhythm (Body Clock)

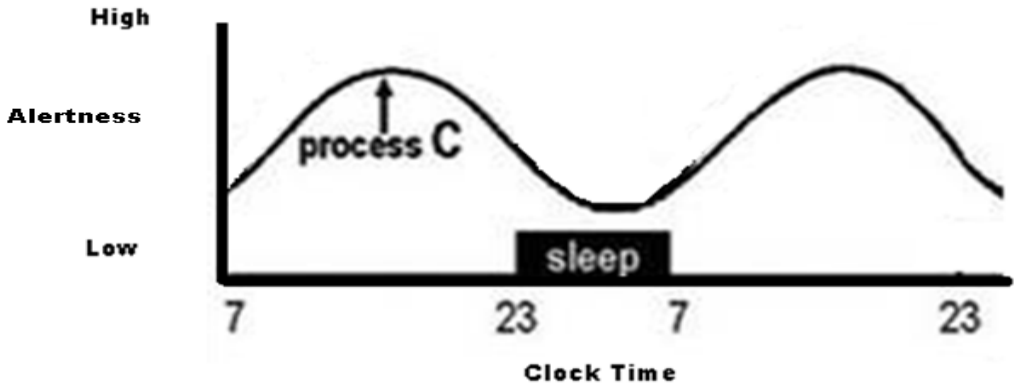
The Circadian Rhythm (Body clock) is an internally generated biological rhythm that allows a number of processes to rise and fall over the twenty four hour period. Commonly, its effects are mostly realised with jet lag when one travels through many different time zones rapidly. This is when the circadian rhythm is out of synchrony with the new environment and can take a number of days to go back to normal.

In a good sleeper, who is in-synch with the environment, the circadian rhythm will naturally rise in the early morning, promoting wakefulness and alertness - this is sometimes known as the alerting force. As the day continues, the circadian rhythm will promote wakefulness until it reaches a peak at about midday (see the diagram below showing this rise and fall in alertness). After this time, the circadian rhythm will start to dip. This initial fall is known as the 'post lunch dip' (you may be familiar with greater feelings of sleepiness after lunch) and as a time for a siesta in other cultures. As we continue through the day, the circadian rhythm continues to fall and does not promote as much arousal as before. With the onset of bedtime and sleep, the circadian rhythm drops to the lowest level and helps to maintain sleep. In this way, sleep pressure is very high, while the alerting effect of our body clock is low, creating the optimal opportunity to sleep.

After this low point, the circadian rhythm will then rise again in anticipation for the next day. The body clock is difficult to manipulate and may be disrupted in a number of poor sleep conditions.

Diagram of Circadian Rhythm (Body Clock)

The diagram below shows how our body clock controls our alertness over the course of the day.



How will this information help me sleep?

It is possible that in individuals with insomnia both sleep pressure and the body clock are not functioning optimally, and bed restriction aims to 'jump-start' these two processes to help promote consolidated and good quality sleep. Bed restriction involves:

(1). Reducing the amount of time that you spend in bed. This will lead to a build-up of sleep pressure to encourage falling asleep straight away and prevent middle-of-the-night awakenings.

(2). Standardising the time you go to bed at and get up at each day. This will help create a strong circadian rhythm, with natural peaks and troughs of alertness similar to a good sleeper.

Implementing Bed Restriction Therapy

Bed Restriction Therapy begins with a one week sleep diary. This is filled out every morning for seven days, recording on the morning of the first night and is to be repeated everyday throughout this first week. This will give an indication of the amount of time you spent awake in bed and how much sleep you actually obtain on an average night – this is often hard to know because your sleep can vary from night to night. Understanding how much time you spend in bed and how much time you spend asleep will help form the foundations of Bed Restriction Therapy.

Working out your average sleep time

The first step in Bed Restriction Therapy is to work out your average sleep time from the sleep diary. **This is found in part 9 of your sleep diary**. By using your sleep diary record from the past seven nights it will be possible to calculate your average sleep time for those nights.

For Example:

Day	Amount Slept	(Minutes)
Monday	6 hours 30 min	(390)
Tuesday	3 hours 30 min	(210)
Wednesday	5 hours 15 min	(315)
Thursday	6 hours 15 min	(375)
Friday	4 hours	(240)
Saturday	6 hours	(360)
Sunday	5 hours 30 min	(330)

Therefore in this example, by adding up the total amount of time slept on average each night and by dividing that by the number of nights you have data for (in this case seven) we are able to calculate your average sleep time.

Total amount slept = 37 hours or (2220 minutes)
÷
Number of days (7)

Average Sleep Time = 5 hours 17 minutes or (317 minutes)
= 5 hours 15 minutes, to the nearest 15 minutes

This example suggests that this person has slept approximately on average five hours and 15 minutes. However, in order to try and get to sleep this person has been spending an awful lot of time in bed in order to obtain this amount of sleep. A more efficient strategy used within Bed Restriction Therapy is to try to match your time in bed to the time that you spend asleep on average. As a result, we shall now attempt to get your sleep into a regular and efficient pattern. This will seek to utilise the two processes that regulate sleep - the previously mentioned circadian rhythm and the accumulation of sleep pressure.

Getting your sleep into a regular pattern

The first modification to your sleep within Bed Restriction Therapy is to set a morning rising time, a time to get out of bed. It is best to calculate a time to rise from bed in the morning as this is something that we can control. In this example, it was decided that due to work reasons the set morning rising time would be at 6 am. We call this the Set Rising Time or Anchor Time as this should be the time that you rise from bed everyday of the week (even weekends!).

With this new set Rising Time, we then work backwards around the clock with the amount of time that you on average slept for the preceding week (previously worked out to be five hours and 15 minutes from the sleep diary). This enables us to decide on your 'Threshold Time'.

The Threshold time is the time after which you can now retire to your bedroom and to bed. In this example, with a Set rising time of 6am and an average sleep time of 5 hours and 15 minutes the Threshold Time is 00:45am; after this you can go to bed.

You can adjust your preferred threshold and rising times e.g. threshold 01:45, then rising time would be 7am. Or threshold 11:45, then rising time would be 5am. This period between going to bed and rising from bed is called the 'sleep window'.

However, it is vitally important to keep a minimum of 5 hours of time spent in bed.

For example, if your average total sleep time is equal to 4 hours and 30 minutes then the minimum of 5 hours of time spent in bed must be applied. With this example the set rising time would be 7:00 am and the threshold time would be 7:00am – **5hours** and therefore equal 2:00am.

Sleep Efficiency

One of the main goals of bed restriction therapy is to make your sleep more efficient. What we mean by this is that, when you go to bed, you fall asleep quickly and have very little wake time during the night. Sleep efficiency therefore refers to the amount of time that you spend asleep as a percentage of the time that you spend in bed. The example below shows how this can be calculated.

Go to bed: 11:00pm
Get to sleep: 12:30am
Wake up: 6:30am
Time asleep: 6 hours (or 360 minutes)
Time in bed: 7.5 hours (or 450 minutes)
Sleep efficiency = time asleep ÷ time in bed (x 100)
= 6 ÷ 7.5 x 100 = 80%

We will use the Sleep Efficiency ratio to help find out changes in your sleep pattern. A 'good sleep efficiency' is around 90% however this can be variable even in a good sleeper. Sleep efficiency scores less than this suggest a poor sleep.

You can work out your average sleep efficiency for the previous week by using your sleep diary. This will give you an indication of how much time you are sleeping when you are in your bed. Here are the steps to work this out:

Sleep Diary

Below is an example of a sleep diary with the parts in red which are used to calculate the time spent asleep on average (number 9) and the average total time in bed (numbers 2 & 4).

MEASURING THE PATTERN OF YOUR SLEEP	DAY 1	DAY 2	DAY 3	DAY 4	DAY 5	DAY 6	DAY 7
1. Did you nap at any point yesterday? If yes, how long for (minutes)?							
2. At what time did you rise from bed this morning?							
3. What time did you wake up at this morning?							
4. At what time did you go to bed last night?							
5. What time did you switch off the light at intending to go to bed?							
6. How long did it take you to fall asleep (minutes)?							
7. How many times did you wake up during the night?							
8. How long were you awake during the night (in total)?							
9. About how long did you sleep altogether (hours/mins)?							
10. How much alcohol did you take last night?							
11. Did you take sleeping pills to help you sleep last night? If so, how many?							

Aim: To sleep for at least 90% of the time you spend in bed

Step 1: Work out your current average sleep

Step 2: Decide on a morning rising time

Step 3: Work out your threshold time

Step 4: Calculate current sleep efficiency

Sleep efficiency is also a useful tool to use when you leave the treatment intervention as it allows you to re-take control of your sleep-wake schedules and implement it when you require it.

Making these changes

You may find that the mornings will provide you with new opportunities to do exercise or catch up with things that you have long put off. It is also beneficial to get as much light as

possible in the morning to help synchronise your internal Circadian rhythm to the morning rising time.

Bed Restriction Therapy also stops negative associations about the bedroom environment - as it strengthens the idea that the bedroom should only be used for sleeping or intimacy. With this therapy you should no longer worry about trying to get to sleep in the bedroom. Instead you will relish going to bed and it will now become a place for you to finally fall asleep.

Questions and Answers

When will I start to get more sleep?

- Once you are sleeping 90% (sleep efficiency) of your time in bed. After this we will be able to Increase your time in bed by 15 minutes.
- You will then be asked to stay with this new pattern for at least another week.
- We will review your progress and make changes each week for the next 4 weeks.

What do I do with this extra evening time?

- Continue with normal daily tasks, whatever you want to do.
- But do not fall asleep before 'threshold time'
- Safety: do not take risks.

Can I nap during the day?

- Napping may reduce the effectiveness of bed restriction therapy.
- Only take a short nap (15-20 minutes) if you are struggling to stay awake during the day.

Total Sleep Time (TST)

Tine in Bed (TIB)

HOURS	3	3.5	4	4.5	5	5.5	6	6.5	7	7.5	8	8.5	9	9.5
3	100													
3.5	86	100												
4	75	88	100											
4.5	67	78	89	100										
5	60	70	80	90	100									
5.5	55	64	73	82	91	100								
6	50	58	67	75	83	92	100							
6.5	46	54	62	69	77	85	92	100						
7	43	50	57	64	71	79	86	93	100					
7.5	40	47	53	60	67	73	80	87	93	100				
8	37	44	50	56	63	69	75	81	88	94	100			
8.5	35	41	47	53	59	65	71	76	82	88	94	100		
9	33	39	44	50	56	61	67	72	78	83	89	94	100	
9.5	32	37	42	47	53	58	63	68	74	79	84	89	95	100

Chart A
Sleep efficiency (SEFF)

Time to Bed

Time to Rise

	03:00	02:30	02:00	01:30	01:00	12:30	12:00	11:30	11:00	10:30
08:30	5.5	6	6.5	7	7.5	8	8.5	9	9.5	10
08:00	5	5.5	6	6.5	7	7.5	8	8.5	9	9.5
07:30		5	5.5	6	6.5	7	7.5	8	8.5	9
07:00			5	5.5	6	6.5	7	7.5	8	8.5
06:30				5	5.5	6	6.5	7	7.5	8
06:00		***Chart B*** Sleep window options			5	5.5	6	6.5	7	7.5
05:30						5	5.5	6	6.5	7
05:00							5	5.5	6	6.5

Appendix 14: Sleep Diary[3]

SLEEP DIARY

Name_____ **Day 1:**_____ Session 4_____

The sleep diary is designed to provide a record of your sleep pattern as well as how you feel on awakening. Please complete one column of the **AM PAGE each morning**, soon after you wake up and the **PM PAGE every evening** before you go to bed. Take a few minutes to do this, trying to be as accurate as you can. It is your best estimate that we are looking for, but try not to get into the habit of clockwatching at night.

PM PAGE: Tonight's sleep

I. Write down here what you have agreed with your therapist to change for this week

MEASURING YOUR TREATMENT PATTERN	DAY 1	DAY 2	DAY 3	DAY 4	DAY 5	DAY 6	DAY 7
1 How ready are you to do this tonight? 0 1 2 3 4 not at all moderately very							
2. How important is it for you to do this tonight? 0 1 2 3 4 not at all moderately very							
3. How confident are you to do this tonight? 0 1 2 3 4 not at all moderately very							

[3] The PM page and first line of the AM page were added to the baseline version (which only contained questions 1-11; and sleep quality questions on the AM page). This sleep diary was given to individuals once SRT started (week 1). The added questions will be analysed with a time-series analysis, but because this was secondary analysis, have not been included in this thesis.

AM PAGE: Last night's sleep

MEASURING THE PATTERN OF YOUR SLEEP	DAY 1	DAY 2	DAY 3	DAY 4	DAY 5	DAY 6	DAY 7
Write down here what you have agreed with your therapist to change for this week How well/often did you do this last night? 0 1 2 3 4 Not very well very well							
1. Did you nap at any point yesterday? If yes, how long for (minutes)?							
2. At what time did you rise from bed this morning?							
3. What time did you finally wake up at this morning?							
4. At what time did you go to bed last night?							
5. What time did you switch off the light at intending to go to sleep?							
6. How long did it take you to fall asleep (minutes)?							
7. How many times did you wake up during the night?							
8. How long were you awake during the Night because of these awakenings (in total minutes)?							
9. About how long did you sleep altogether (hours/mins)?							
10. How much alcohol did you take last night?							
11. Did you take sleeping pills to help you sleep last night? If so, how many?							

MEASURING THE QUALITY OF YOUR SLEEP

1 How rested / restored do you feel this morning? 0 1 2 3 4 not at all moderately very							
2. How alert do you feel this morning? 0 1 2 3 4 not at all moderately very							
3. How would you rate the overall quality of your sleep last night? 0 1 2 3 4 poor very good							

Appendix 15: Bed Restriction Specific Adherence Scale

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Appendix 10-Adherence Scale
NAME:_____ Date: _____Session 1_____

Bed Restriction therapy

How often was each of the following statements true for you during the past week? Please Circle the most accurate response.

	None of the time	A little of the time	Some of the time	A good bit of the time	Most of the time	All of the time
I followed the bed restriction programme…	1	2	3	4	5	6
I went to bed at my calculated 'threshold time' on..:						
(a) weekdays	1	2	3	4	5	6
(b) weekends	1	2	3	4	5	6
I got up at my calculated 'rising time' on...:						
(a) weekdays	1	2	3	4	5	6
(b) weekends	1	2	3	4	5	6

Appendix 16: Mental Arithmetic Task

Digit Span:
In this task I will read you a number sequence, starting with 2 digit. I will ask you to recite them in correct order.

The number sequence increases after each correct response by one digit. This is continued until the individual makes two consecutive mistakes. This task can also be done in reverse: the individuals recite the given sequence in reverse.

```
4 7
1 6 2
4 7 8 8
8 6 1 2 6
3 2 1 9 6 4
9 4 2 4 5 3 5
9 1 1 3 6 6 8 6
3 8 9 9 2 4 9 6 8
9 2 8 7 7 3 2 6 5 5
9 4 3 4 3 5 6 8 3 3 2
1 6 6 7 2 8 1 9 6 5 7 5
```

Number Task:
I am going to ask you to countdown from the number 300 in steps of 3.

300	450	1736
297	443	1728
294	436	1720
291	429	1712
288	422	1704
285	415	1696
282	408	1688
279	401	1680
276	394	1672
273	387	1664
270	380	1656
267	373	1648
264	366	1640
261	359	1632
258	352	1624
255	345	1616
252	338	1608
249	331	1600
246	324	1592
243	317	1584
240	310	1576
237	303	1568
234	296	1560
231	289	1552
228	282	1544
225	275	1536
222	268	1528
219	261	1520

Appendix 17: Patient Characteristics of Sample Recruited

N	30
Age (mean yrs)	48 (range 19-65)
Gender (female)	23 (77%)
Education (mean yrs)	15 (range 11-20)
Insomnia subtype (n)	
Onset	9
Maintenance	8
Mixed	8
Early morning awakening	2
Insomnia duration (mean yrs)	20 (SD 19; 7 with childhood onset)

Appendix 18: Correlation between Adherence and Outcome

Table 26: Spearman Rho correlations between Adherence Variables and Outcome post-treatment and follow-up

	ISI Change		Diary Sleep Efficiency Change		Sleep Diary SOL		Sleep Diary WASO	
	Posttreatment	Follow-up	Posttreatment	Follow-up	Posttreatment	Follow-up	Posttreatment	Follow-up
Questionnaire								
Total score	-.25	-.12	.19	.05	-.20	-.23	.00	.15
Weekdays	-.32	-.23	.19	.15	-.03	-.10	-.03	.07
Weekends	-.19	-.05	.19	.15	-.29	-.30	.00	.17
Rising time	-.27	-.16	.18	.14	-.05	-.10	-.01	.14
Threshold	-.16	-.04	.19	.05	-.32	-.30	.06	.21
Sleep Diary								
Composite	.20	.20	-.23	-.30	-.05	.09	.25	.23
Rise Deviation Variance	.15	.29	-.25	-.35	-.09	.02	.22	.13
Threshold Deviation Variance	.33	.07	-.17	**-.35** [+]	-.16	.05	.28	.27
Actigraphy-Marker								
Composite (MarkCOMP)	.24	.23	**-.49***	**-.40***	.10	.19	.18	.19
Rise Deviation Variance	.28	.35	-.33	**-.38** [+]	-.14	-.05	.19	.16
Threshold Deviation Variance	.23	.02	-.32	-.32	.05	.12	.15	.18
Actigraphy-Movement								
Composite	.26[a]	.08[a]	-.21[a]	-.18[a]	.06	.12	.11	.05
Rise Deviation Variance	.26[a]	.15[a]	-.21[a]	-.14[a]	-.08	-.04	.12	.11
Threshold Deviation Variance	.18[a]	-.05[a]	-.25[a]	-.09[a]	.21	.27	-.07	-.01

*Significant correlation, $p<0.05$; [+] Trend at $p \leq 0.07$ [a] Pearson's r for normally distributed data

Appendix 19- Mann-Whitney test: non-signifcant results comparing motivation scores across stages of change

Table 27: Mann-Whitney test comparing motivation scores across stages of change

	Median C/Pr [IQR]	Median Ac/M	p-value
Motivation Week 2	9.8 [9.1-10]	9.9 [9.6-10]	0.78
Motivation Week 4	9.0 [8.3-10]	9.9 [9.3-10]	0.38
Motivation FU	9.3 [9.0-9.7]	9.7 [9.4-10]	0.13

C=comtemplation; Pr=Preparation; Ac= Action; M=Maintenance; IQR= Interquartile range

Appendix 20- Spearman Correlations between baseline variables and adherence

Table 28: Spearman Correlations between baseline variables and adherence

Baseline Questionnaire	Spearman's rho	P-value
Baseline IPQ-R timeline subscale[4]	0.09	0.65
Baseline IPQ-R timeline cyclical	0.04	0.85
Baseline IPQ-R IPQ Consequences	0.12	0.54
Baseline IPQ-R Personal Control	-0.1	0.60
Baseline IPQ-R Treatment Control	0.08	0.68
Baseline ESS	0.03	0.89
Baseline FSS	0.13	0.51

[4] The identity and causal items are not included here. Identity was not measured to a) reduce the item numbers and b) because it did not seem appropriate for a disorder with such different symptoms (sleep onset vs. sleep maintenance). There is not a total score for causal attributions, thus although measured, this was not analysed here.

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