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**‘Breaking Good News’: Neurologists’ experiences of discussing SUDEP with patients
in Scotland**

And Clinical Research Portfolio

Volume 1
(Volume 2 bound separately)

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Submitted in partial fulfilment of the requirements for the degree
of
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Systematic Review

A systematic review of the qualitative literature examining the experience of being diagnosed with epilepsy.

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Abstract

A meta-ethnography approach synthesised themes found in the qualitative literature examining the experience of being diagnosed with epilepsy. Eleven studies were assessed and five super-ordinate themes emerged of 'initial onset', 'diagnosis complications', 'information sources', 'time to process', 'adjustment and role change' and 'relationship with healthcare staff'. 'Relationship with healthcare staff' affects all the super-ordinate themes apart from 'initial onset'. Moreover, patients value the relationship in its own right and the importance of this relationship is highlighted and discussed. The results would support interventions that aim to increase the awareness of epilepsy signs amongst hospital and school staff and adds to the literature suggesting relationships form a key part in understanding patients' adjustment to epilepsy.

Keywords: Epilepsy, diagnosis, meta-ethnography, qualitative, patient experience

Highlights

- *Qualitative literature examining patients' experiences of being diagnosed was systematically reviewed and synthesised using a meta-ethnography approach*
- *Research from children, young adults, older adults, and relatives of people with epilepsy was assessed and a typical experience of being diagnosed with epilepsy is presented. Complications and delays with diagnosis are commonly reported*
- *The relationship with healthcare provider is of central importance to the experience of being diagnosed with epilepsy*

1. Introduction

The experience of living with epilepsy has been explored using a wide range of qualitative approaches such as a latent content analysis [1], a phenomenological approach [2], and an ethnographic approach [3]. Researchers have been interested in the experiences of children with epilepsy [4], their parents [5], adolescents with epilepsy [1], young adults with epilepsy [6], and older adults with epilepsy [7]. Kerr, Nixon, and Angalakudit [8] conducted a systematic review of the qualitative literature looking at the impact of epilepsy on patients' lives. The authors reviewed 18 qualitative studies and devised a conceptual model of 23 factors which they felt influenced the lives of children and adults with epilepsy. Their model suggested that some factors directly affected patients' lives (such as the physical effects of epilepsy) while other factors had an indirect impact (e.g. a financial impact for adults due to the medication cost). The authors also stated that many of the factors were shared amongst children and adults.

Studies have found the experience of being diagnosed with epilepsy is important for patients however this has typically been explored via a broader qualitative investigation of epilepsy. For example, Sample et al. [9] found that the experience of being diagnosed was an important aspect when patients and their families considered how they had found support. Tonberg et al. [6] examined the experiences of young adults with regards to their knowledge of Sudden Unexpected Death in Epilepsy (SUDEP) and found that participants wanted this information but only after the initial diagnosis of epilepsy. These studies suggest that the experience of being diagnosed has an impact on how people with epilepsy gain support and knowledge for the condition.

A smaller number of studies have focused solely on the experience of being diagnosed with epilepsy. Miller, Buelow, and Bakas [7] examined older adults' experience of being

diagnosed with epilepsy and why there are often delays in diagnosing in this population. They found themes of ‘being dismissed’ and ‘frustration’ typically characterised their experiences. Two qualitative examinations have examined the experiences of parents following their child’s diagnosis with epilepsy [5, 10]. Buelow and Shore [10] examined the experience in the context of identifying parental or healthcare ‘failures’ on the way to a successful diagnosis. Nguyen, Pertini and Kettler [5] explored the coping processes of parents following their child’s diagnosis and noted themes of ‘loss of control’ and ‘emotional venting’.

These studies helped identify areas of improvement and helped understand the adjustments people with epilepsy, or their caregivers, face after being diagnosed. As a result of the specific populations examined, however, the general applicability of these findings may be somewhat limited. To the author’s knowledge there has been no attempt to synthesise the literature examining the experiences of being diagnosed with epilepsy.

This systematic review produced a comprehensive account of the experience of being diagnosed with epilepsy by synthesising the qualitative research that has either directly or indirectly examined this experience. The literature was critically appraised and the findings were synthesised to produce third order themes. Using a meta-ethnographic approach [11] a set of themes was produced which explain the experience of an epilepsy diagnosis.

2. Aim

To critically appraise and synthesise the qualitative literature that has examined the experience of being diagnosed with epilepsy.

3. Material and Methods

3.1 Method of synthesis

The themes relating to the experience of being diagnosed with epilepsy were synthesised using a meta-ethnographic approach [11]. This process follows seven steps [12] and allows the translation of studies into one another, meaning that comparison is possible. The extant literature was systemically reviewed to find studies which have qualitatively examined the experience of being diagnosed with epilepsy, or which have investigated the experience as part of a qualitative investigation into epilepsy. Those studies were then reviewed to select papers that provide themes that account for the experience of being diagnosed with epilepsy. Studies were first assessed by the researcher in terms of title and abstract. Those studies which appeared suitable were then reviewed in full and reasons for subsequent exclusion detailed. Included studies were then quality appraised.

By following the meta-ethnographic approach as described by Atkins et al. [12], an overall understanding of the experience of an epilepsy diagnosis was then derived [13]. The main themes that were relevant to the experience of being diagnosed were entered into a table (see Table 1). Themes were then contrasted and compared between studies to create third order themes that represented shared or compatible findings between studies. This process involved exploring the individual themes of one study and comparing it with themes in other studies, in addition to the third order theme being created. The resulting third order theme, although composed of pre-existing themes found in individual studies, expressed something

novel regarding the overall experience of being diagnosed with epilepsy. The third order themes were then analysed to present a model that explained how they may be related to one another (this is known as a ‘line-of-argument’ synthesis [12]).

3.2 Reflexivity

The researcher was conducting qualitative research regarding clinicians’ views of discussing SUDEP at the time of this meta-ethnography.

3.3 Search strategy

MEDLINE (1990-present), EMBASE (1990 – present), PsychINFO (1990-present), and Web of Science (Core Collection 1990 – present) were searched using the EBSCO Host. The search was conducted on the 10th February 2016.

The following search terms were separated by Boolean operators:

1. epilepsy, OR seizures, OR tonic-clonic

AND

2. diagnos*, OR patient* experience*, OR patient* perspective*, OR patient* reaction*, OR personal experience

AND

3. Qualitative, OR qualitative analysis, OR qualitative research, OR content analysis, OR thematic analysis, OR grounded theory, OR Interpretative Phenomenological Analysis

Search terms varied to accommodate specific databases (see Appendix 2 for the search terms used for each database).

3.4 Results of Search

486 citations were produced as a result of using the above search terms. 169 of these were duplicates. The remaining 317 articles were assessed by title, abstract and full text according to the following inclusion and exclusion criteria:

3.5 Inclusion Criteria

- Studies that directly examined the experience of being diagnosed with epilepsy. ‘Being diagnosed’ referred to that process, or period of time, when an individual notices symptoms, seeks medical investigation, is informed of their condition and given information or advice regarding it, the subsequent immediate adjustment (<6 months following diagnosis), and thoughts on the process. Included studies could investigate any part of this process
- Studies were considered if they related to diagnosis as experienced by patients, caregivers, family members or partners
- Any study that used a qualitative interview and analysis to examine patient experiences of an epilepsy diagnosis. This included focus groups
- Studies that examined all age ranges were considered
- Studies which contained at least one theme relating to being diagnosed with epilepsy. The theme had to be explored in an independent section of the Results

or Analysis section (i.e. it was not sufficient if the theme was a sub-theme of another theme)

- Studies were included if the experience of being diagnosed occurred when a medical professional gave a diagnosis of epilepsy as a result of a neurological disorder (i.e. those studies which investigated experiences of people who were diagnosed with epilepsy as a result of ‘spirits’ were excluded)
- Published in a peer reviewed journal
- Published in the English language

3.6 Exclusion Criteria

- Studies that did not include a qualitative analysis
- Studies which analysed the experience of living with a diagnosis of epilepsy (for inclusion they had to relate to the diagnostic process or experience)
- Studies that explored the diagnosis of Non-Epileptic Seizures (NES) were not included due to the additional themes that are present for this population (such as ‘resisting the diagnosis’ [14])

The results of this process are shown in figure 1.

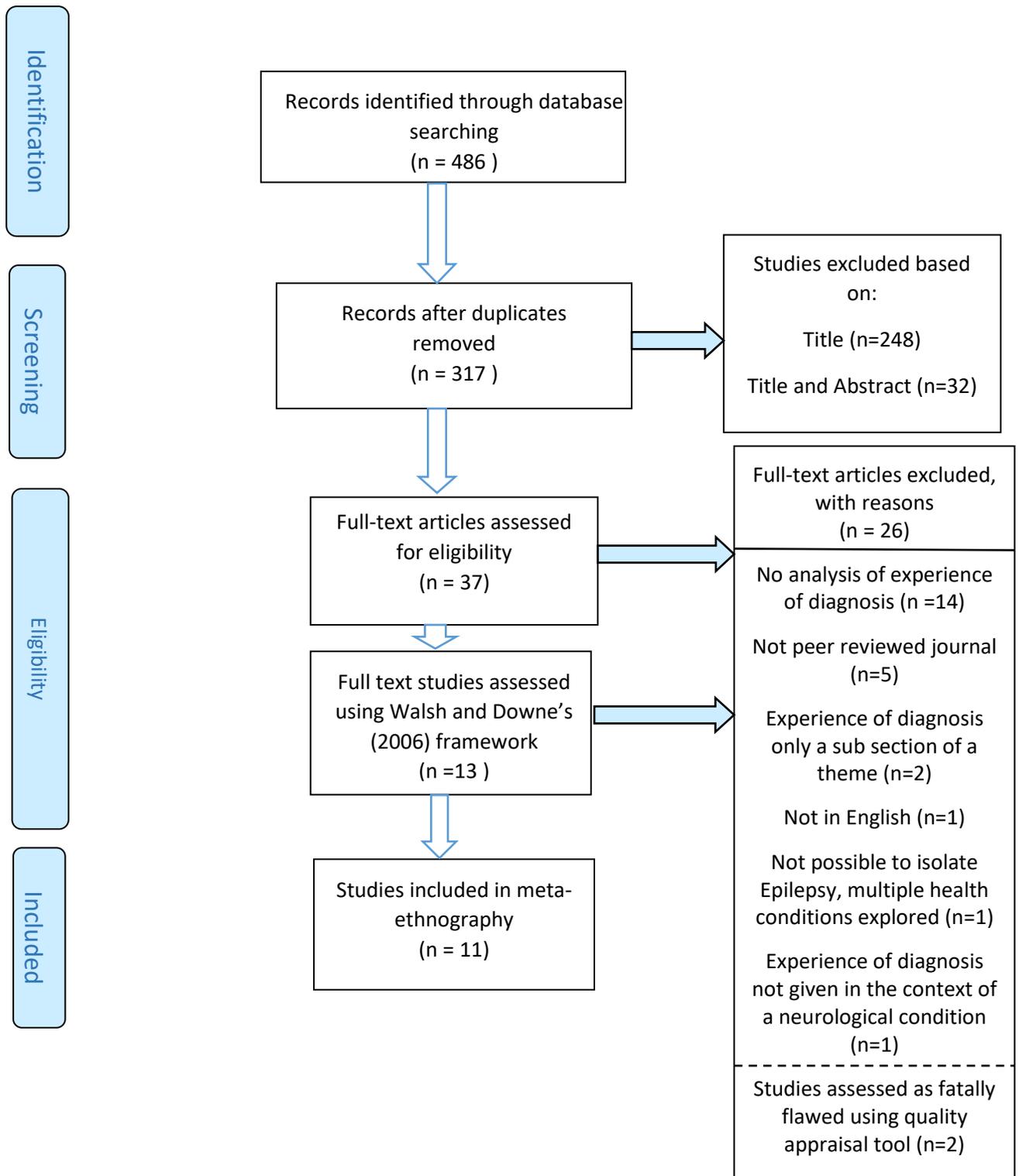


Figure 1: Flowchart of search results

4. Results

4.1 Quality appraisal

While it is recognised that a checklist approach can be reductionist when quality appraising qualitative literature [15], Walsh and Downe [16] produced a checklist of quality criteria after assessing a range of critical appraisal materials. Dixon-Woods et al. [17] also suggest five questions to assess if studies are ‘fatally flawed’. All studies were firstly appraised using the Walsh and Downe [16] checklist (See Appendix 3). The 8 studies which did not satisfy all 12 quality indicators were then assessed using the Dixon-Woods et al. [17] questions to ensure that potentially insightful analyses were not excluded on the basis of minor methodological flaws (see Appendix 4). Two papers [18, 19] were excluded from the review on the basis of being fatally flawed (see Appendix 4).

All of the included studies gave a clear rationale for the research. None of the studies detailed a systematic approach to assessing relevant literature however, Sample et al. [9] included an in depth literature review justifying their phenomenological approach. Most of the studies did not justify why they chose their qualitative methodology. The exception to this, again, was the Sample et al. [9] paper.

It was common for papers to employ multiple researchers for coding and the analytic approach was deemed appropriate for most papers assessed. The decision trail of all papers was clear to follow. All papers used data to support their findings however the Miller, Bakas and Buelow [20] paper did not provide sufficient quotes to support interpretations.

Sample et al. [9] discussed the relationship between the researcher and participants. No other paper mentioned researcher reflexivity, however, there was evidence that papers had considered researcher influence on the research process and demonstrated self-awareness [4-7, 21].

All papers demonstrated sensitivity to ethical issues and noted confidentiality procedures however it was felt that 2 papers could have considered providing support for participants who may have been distressed discussing their experiences [4, 22]. All papers discussed how their findings may contribute to existing theories and literature.

Three of the studies were randomly selected for quality review and double rated by an independent reviewer. Co-rater ratings are shown in Appendix 3. Disagreements in rating were discussed and agreement reached by discussion.

The characteristics of studies included in the meta-ethnography are shown in Table 1.

Table 1: Characteristics of studies

Study (year) Country	Method of analysis	Participants	Article themes relating to diagnosis
Baca et al. (2015) [23] USA	Thematic analysis	37 individual interviews with parents (mean age = 41.3 (SD 7.3)) of children with epilepsy who had undergone resective epilepsy surgery (mean age at surgery 8.2 (SD 4.7))	Theme 1: Recognition 'something is wrong'. Theme 2: Searching and finding: 'a journey around the world and very circuitous'
Buelow and Shore (2006) [10] USA	Cross case content analysis	21 individual interviews with parents of children (ages 8-16) with epilepsy and significant learning problems	Theme 3.1: The ideal trajectory of diagnosis. Theme 3.2: Failures in recognition and treatment
Harden et al. (2015) [21] β Scotland	Inductive thematic analysis	27 individual interviews with young adults (aged 18-29) with epilepsy	SUDEP information giving and seeking. SUDEP information giving and reported behavioural change
Tonberg et al. (2015) [6] β Scotland	Inductive thematic analysis	27 individual interviews with young adults (aged 18-29) with epilepsy	Theme 2: Knowing about SUDEP Theme 3: Impact of being told about SUDEP
McNelis et al. (2007) [4] USA	Qualitative focus group design	4 focus groups were held with 2 groups of children (6 children with seizures aged 7-14; 5 children aged 9-15) and 2 groups of parents (6 mothers and one father; 6 mothers and two fathers; ages not specified)	Difficulties/struggles/problems. Need for information. Fears and concerns

Miller, Buelow and Bakas (2014b)* [7] USA	Content analysis	20 individual interviews with older adults (aged 60-80) with epilepsy	Theme 1: Delayed diagnosis Theme 2: Non-delayed diagnosis
Miller, Bakas and Buelow (2014a)* [20] USA	Content analysis	20 individual interviews with older adults (aged 60-80) with epilepsy	Theme 1: Information Theme 5: Commitments Theme 6: Relationships
Nair, Jack and Strohm (2016) [24] Canada	Directed content analysis	27 individual interviews with bereaved relatives (parent, sibling, spouse, or child aged at least 18 years) of a person with epilepsy (18 were parents of children with epilepsy)	Opinion on whether to discuss SUDEP. Method and timing of SUDEP discussion
Nguyen, Pertini and Kettler (2015)[5] Australia	Theory driven thematic analysis	21 individual interviews with mothers of a child (aged 3-12) diagnosed with epilepsy (diagnosis at least 6 months prior, but no longer than 2 years prior to interview)	The adjustment process. Cognitive appraisals. Coping behaviours
Risdale, Kwan and Morgan (2003) [22] England	No specific approach stated	22 individual interviews with patients (aged 17-83) who had attended a nurse run epilepsy clinic	Challenges for patients
Sample et al. (2006) [9] USA	Phenomenological approach	4 focus groups comprising 31 participants (7 aged 0-21, 24 aged 22-64), either people with epilepsy or family members	Theme 1: Searching, hoping, finding, trying, searching

*Same sample studied, different focus of study,

β Same sample studied, same focus of study, alternative conclusions and themes considered.

The article themes relating to diagnosis were then expanded and unpacked in a subsequent column – noting specific components of the article theme and illustrative direct quotes from participants. Discussion based themes that related to the article themes were also noted (Atkins et al. [12] refer to these as ‘2nd order themes’). Article and discussion themes were compared across papers and through the process of constant comparison, third order themes were developed. An illustrative example of this is shown for one of the papers in Appendix 5.

The meta-ethnography produced the following third order themes: 1) Initial onset 2) Diagnosis complications 3) Relationship with Healthcare Staff 4) Information (sources, content and ownership) 5) Time to Process the Diagnosis 6) Role Change and Adjustment. The contribution of individual papers to the generated themes is shown in table 2. Each theme is discussed in detail below.

4.2 Initial Onset

The experience of being diagnosed with epilepsy begins by noticing the symptoms of epilepsy. Initially, patients may not seek medical attention as symptoms do not interfere with their daily activities or work [7].

‘I was still working at the time...it was low on my to-do list’ [7, p6]

Some of the same study’s participants were embarrassed about their symptoms and only sought medical attention when their relatives persuaded them:

‘my daughter thought I was having a stroke...finally they convinced me to go’ [7, p7]

The above quote highlights another feature of the initial onset in that there is often confusion about symptoms and what they are experiencing, however, there is a recognition that something is wrong, and it is serious [9, 23]:

'I knew there was something wrong with me. I did not know what.' [9, p653]

This recognition is shared by relatives:

'I didn't know what was going on and I had no idea. I just knew it was an emergency' [23, p825]

'...he started putting his head back. I thought he was tired and I laid him on the couch and he had a grand mal seizure' [10, p442]

Buelow and Shore [10] identify that the epilepsy symptoms of their participants' children were often first identified by a healthcare professional or teacher.

Table 2: Contribution of papers to 3rd order themes

	Initial Onset	Diagnosis Complications	Relationship with Healthcare Staff	Information (sources, content and ownership)	Time to Process the Diagnosis	Role Change and Adjustment
Baca et al. (2015) [23] <i>USA</i>	Recognition – something is wrong		Searching and Finding		Searching and Finding	Searching and Finding
Buelow and Shore (2006) [10] <i>USA</i>	The ideal Trajectory Failures in recognition and treatment	The ideal Trajectory Failures in recognition and treatment	Failures in recognition and treatment			
Harden et al. (2015) [21] β <i>Scotland</i>			SUDEP information giving and seeking	SUDEP information giving and seeking	SUDEP information giving and seeking	SUDEP information giving and seeking
Tonberg et al. (2015) [6] β <i>Scotland</i>			Knowing about SUDEP	Knowing about SUDEP	Knowing about SUDEP Impact of being told about SUDEP	
McNelis et al. (2007) [4] <i>USA</i>		Difficulties/struggles/pr oblems	Difficulties/struggles/pr oblems	Difficulties/struggles/pr oblems	Need for information Fears and concerns	Difficulties/struggles/pr oblems Need for information Fears and concerns

Miller, Buelow and Bakas (2014)* [7] USA	Delayed diagnosis Non-delayed diagnosis	Delayed diagnosis Non-delayed diagnosis				
Miller, Bakas and Buelow (2014)* [20] USA				Information		Commitments Relationships
Nair, Jack and Strohm (2016) [24] Canada			Method and timing of SUDEP discussion	Opinion on whether to discuss SUDEP Method and timing of SUDEP discussion	Method and timing of SUDEP discussion	Opinion on whether to discuss SUDEP
Nguyen, Pertini and Kettler (2015)[5] Australia				Coping behaviours.	The adjustment process	The adjustment process Cognitive appraisals Coping behaviours
Risdale, Kwan and Morgan (2003) [22] England				Challenges for patients	Challenges for patients	Challenges for patients
Sample et al. (2006) [9] USA	Searching, hoping, finding, trying, searching					

*Same sample studied, different focus of study

β Same sample studied, same focus of study, alternative conclusions and themes considered.

4.3 Diagnosis Complications

Unfortunately, it would appear that receiving an incorrect or delayed diagnosis is common and a shared experience across children, adults and older adults [4, 7, 9, 10, 22].

‘The first one [neurologist] misdiagnosed me for 3 years’ [9, p653]

‘Every time I went back they would diagnose me with something different or say it was something I already had...so it took almost a year and a half.’ [7,p6]

This is distressing for patients and their families and in addition to the potential financial cost to American patients [7], it may also result in inappropriate treatment [10]. Parents describe ‘a feeling of desperation to get a diagnosis’ for their child [23, p825].

The reasons for delays are varied but include people with epilepsy and their families being unfamiliar with symptoms [10], being reluctant to seek treatment [7], and healthcare staff being unfamiliar with signs and symptoms of epilepsy [7, 9, 10]. Two studies suggest that those of a lower socio-economic status tend to wait longer for a successful diagnosis [7, 10].

Factors were identified across studies that lead to a more timely diagnosis. Children under the care of a physician for another medical condition were identified as receiving a successful diagnosis [10] and patients who exhibit more classical symptoms of seizures were more readily diagnosed than those who exhibit more ambiguous signs [7].

4.4 Relationship with Healthcare Staff

The relationship with healthcare staff is of fundamental importance to the experience of being diagnosed with epilepsy and the majority of studies included themes or quotes that reflected this [4, 6, 9, 10, 21, 23, 24]. During the initial stages of the diagnosis, some patients

feel that healthcare staff are dismissive of their concerns or do not believe their descriptions [4, 7, 9, 10, 23]:

'I didn't say anything to the doctors or anything at that point because I [didn't think] I'd be believed. [I thought] I'd just be [considered] a hysterical mother.'
[10, p443]

'He said my symptoms had to be hormonal...and the last time I saw him he said maybe I was...cursed' [7, p6]

'The doctors questioned whether or not I was making this up' [9, p653]

Staff may be reluctant to form relationships:

'I had difficulty talking to the doctor and nurses; I did not feel part of the team; the doctors and nurses never talked to my child either' [4, p198]

In contrast, patients note how helpful a positive relationship with healthcare staff can be to the experience of being diagnosed:

'The seizure clinic down at [a specific hospital] ...I can call 24 hours a day, and they have somebody there to answer my questions. And that helps a lot.' [9, p654]

Additionally, patients and family members note that the relationship with the neurologist, physician or epilepsy nurse specialist determines who should relay information about SUDEP risk [6, 21, 24]. And that it is important that clinicians discuss SUDEP because if not:

'it could create a bit of mistrust' [21, p234]

Trust appears to be a crucial component of the relationship formed with professionals:

'the big thing is finding somebody who you can trust, who you're comfortable with, who [the child] is comfortable with, who will respond to your questions...' [4, p198]

4.5 Information (sources, content and ownership)

Two studies suggested that people can find it difficult to get enough information about epilepsy [4, 20] and this may cause embarrassment when explaining the condition to others [20]. People with epilepsy may also forget to ask important questions when speaking to their clinicians [22].

Information is valued most from people, rather than leaflets, books or the internet [20, 21, 24]. This is because an 'open dialogue can occur and that questions can be addressed' [24, p23]. Information from books or the internet may also be confusing for older adults with epilepsy [20]. People commonly search for information after being diagnosed however, and the internet can be a useful resource for this [9]. Information is appreciated when it comes from other people with epilepsy, or parents of children with epilepsy [5, 20]. This is reported to be 'informative and reassuring' [5, p32] and the same researchers suggest these opportunities should be facilitated. McNelis et al. [4] note the contribution that general health professionals and school staff make in providing information about epilepsy and suggest that increasing the knowledge of epilepsy amongst these staff groups would be a valuable clinical intervention.

Patients want to be informed about SUDEP at or around the time of diagnosis [6, 21, 24]. Moreover, patients feel they have a right to this information.

'you have a right to know...I really kind of resent the idea that a doctor in particular would not share that information because he had some personal feelings about 'that's not good for her [to know about SUDEP]' [24, p22]

4.6 Time to Process the Diagnosis

People with epilepsy and their families need time to process the diagnosis [4, 6, 21, 22-24]. This is for two reasons: firstly, there is too much information to take in [4, 23, 24]. Some participants noted it was:

'Like an avalanche of information coming at us' [23, p825]

'I think it's a learning curve process on a need-to-know basis. If you get too much information too fast, you get overwhelmed and confused, you don't know the jargon' [4, p199].

This indicates that learning about epilepsy can be a cognitively demanding process. Secondly, time is required to process the emotional impact of the diagnosis [5, 6, 21, 22]. When asked about when they would like to receive information about SUDEP one participant answered:

'The [session] after [diagnosis]...cos getting epilepsy is a bit of a shock to everybody never mind finding out that you could die from it' [6, p101]

The emotional impact of receiving the diagnosis may be less for older adults than for younger adults:

'I think because all these years I have had to cope with a cardiac problem, it's just something more that I have taken on board, you know' (64 year old woman, [22, p71])

4.7 Role Change and Adjustment

After receiving a diagnosis of epilepsy it is common for people to report a role change with family members. Parents of a child diagnosed with epilepsy often become advocates [4, 5, 23]. Nguyen, Pertini and Kettler [5] describe this as having to ‘relentlessly negotiate for their child’s needs’ [5, p31].

Other role changes were noted:

‘We’ve been together 40 years...real independent. But now...[my husband] hovers.

I went from being wife...to child’ [20, p29]

There is a recognition that parents may become more protective of their child if they learn about SUDEP [24]. Positive role changes are also identified: ‘mothers [take] pride in feeling that they were well placed to offer advice to others’ [5, p29].

Following the initial reaction to the diagnosis, people adjust to the experience of being diagnosed with epilepsy [5, 20, 21]. Some of the older adults in Miller, Bakas and Buelow’s [20] study adjusted to the diagnosis by withdrawing from work or volunteering commitments:

‘I went from part time to really part time’ [20, p28]

In contrast, the parents in Nguyen, Pertini and Kettler’s [5] study indicate that things get better following the diagnosis and the authors identify a number of cognitive strategies which facilitate this change.

‘it does get better...it’s not as traumatic as when it first started’ [5, p27]

Harden et al. [21] note that discussions about SUDEP appear to have little influence on their participants' behaviour and this was a way of adjusting to the knowledge they could die from epilepsy.

Figure 2 shows a diagrammatic representation of the third order themes. The diagram represents the temporal features of the themes identified and the dominance and influence of the 'relationship with healthcare staff' is emphasised.

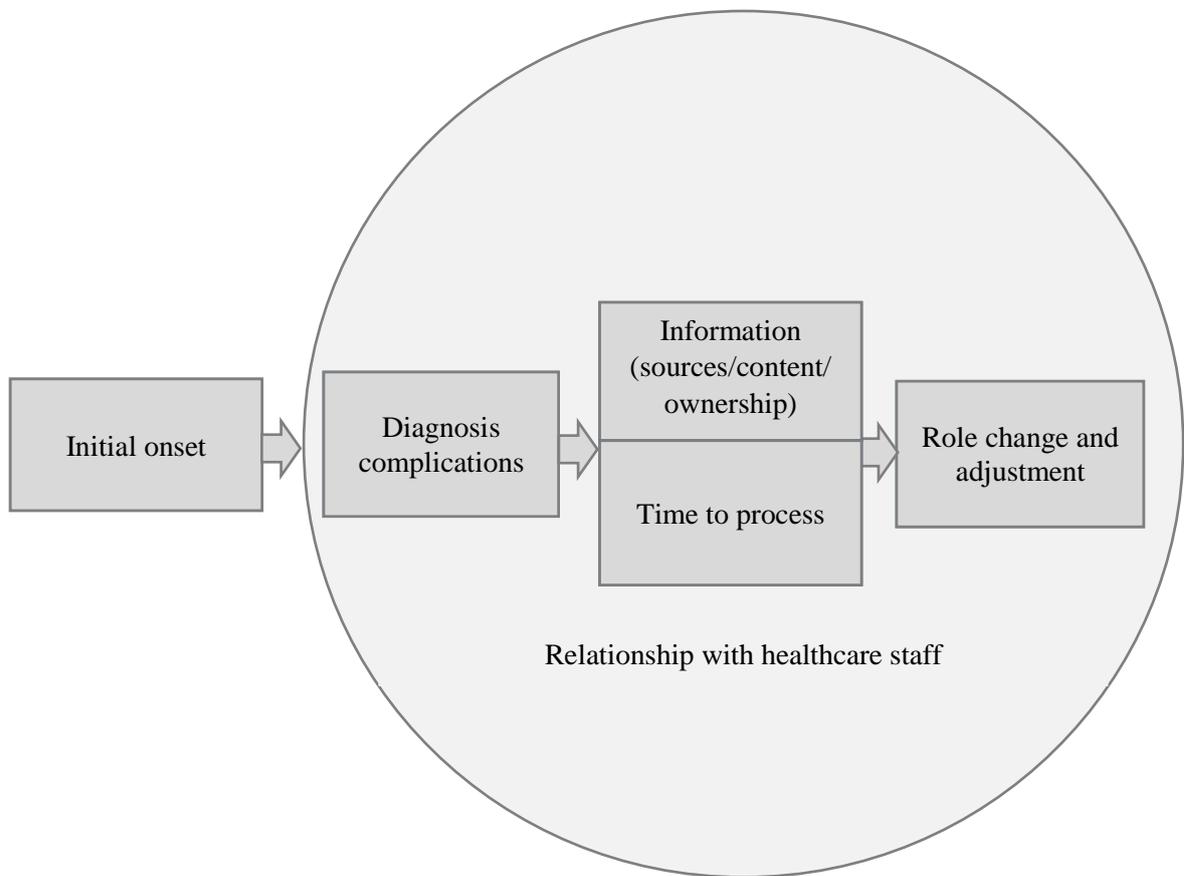


Figure 2: Relationship of the themes

5. Discussion

Some patients do not report initial symptoms of epilepsy either because they are misidentified, or because they fear an epilepsy diagnosis will cause them inconvenience. Consequently, relatives, and healthcare or school staff play an important role in noticing initial epilepsy symptoms and prompting action. The results of this meta-ethnography support the recommendation of McNelis et al. [4] that general healthcare and school staff should be aware of epilepsy symptoms.

The ubiquity of diagnosis complications across age ranges is a prominent theme. While there may be factors that make epilepsy diagnosis with older adults more complicated [7], this meta-ethnography would suggest that complications or delays in diagnosis are not unique to this population.

The information patients receive about their condition is found to be wanting in some respects. Although Risdale, Kwan and Morgan [22] note that patients forget to ask important questions when they see their physician, one may ask if that burden should lie solely with the patient, and what the presence of such cognitions indicates about the quality of the patient-doctor relationship. The fact that patients value information from other patients or relatives (and that they value providing that information) suggests that Nguyen, Pertini and Kettler [5] are correct in suggesting that even informal opportunities to do so should be promoted. Research has indicated that parents report a reduction in overall stress when they have an opportunity to receive information about their child's chronic condition from parents in a similar situation [25]. The results of this meta-ethnography would support this finding and may suggest that a similar desire for support is present in adult patients living with epilepsy.

Time is needed to process the experience of being diagnosed. Tonberg et al. [6] note that the appointment following diagnosis may be appropriate for information regarding SUDEP. This may suggest that as few as two appointments may be sufficient for time to process the information. Time is also required to process the emotional impact of the diagnosis however the impact may be less for older adults.

The role changes following diagnosis were rarely regarded as positive. It may be beneficial to consider what causes some patients to withdraw socially [20] and others to feel that ‘it does get better’ [5, p27].

5.1 Centrality of the Relationship With Healthcare Staff

An important finding of this meta-ethnography has been the centrality of the relationship with healthcare staff to all of the themes relating to the experience of being diagnosed with epilepsy (apart from the initial onset). The relationship appeared to either positively or negatively influence the other themes found. The relationship with healthcare staff often appeared to ameliorate or exacerbate the impact of diagnosis complications and affected patients’ perceptions of the information they received. The relationship with the healthcare staff may also influence the required time to process the diagnosis (informationally and emotionally) and in regards to how patients adjust to their new roles. Given the importance placed on the relationship in other areas of the diagnosis, this may suggest an avenue for intervention.

The relationship with healthcare staff is important not just because it affects the other third order themes of the experience of being diagnosed – it is an important factor in its own right. Patients value the relationship with healthcare staff as a source of trust, understanding and support. This is not a unique finding with regards to how patients cope with chronic

conditions. Previous research has identified that information seeking from healthcare professionals is a coping strategy to gain control over an unpredictable condition [26] and the positive relationship provides an additional sense of support [27]. This systematic review supports these findings and in so doing so, contributes to the literature suggesting people with long term health conditions find great value in the relationship with their healthcare provider.

It is interesting to note the parallels to Kerr, Nixon, and Angalakudit [8] which noted the concerns of patients living with epilepsy were relationships with friends, partners and families. It has been evident for some time that adjustment to chronic illness is a multi-factorial process [28]. This meta-ethnography has highlighted the importance of relationships with healthcare staff as a factor which may exert a strong influence on the experience of being diagnosed with epilepsy and the subsequent adjustment to the condition.

5.2 Limitations

One of the strengths of this study is that studies on unique populations contributed to themes which could be translated to children, adults and older adults (Table 2). This suggests that it is possible to conceive of ‘a diagnosis experience’, despite the variety of individuals and contexts epilepsy presents in. However, it may be useful to explore if the themes identified apply equally to all patient groups. For example, evidence that specific populations commonly wait longer for diagnosis [10] may suggest this is not the case. In a similar regard, particular themes relating to the experience of being diagnosed will have been influenced by the country and culture of the studies involved. The potential cost implications of a delayed diagnosis for American patients has already been noted [7], however, there may be additional

impacts on health care delivery between privatised and public health care systems (e.g. access to resources, or use of care pathways).

It is worth acknowledging the lack of qualitative research focusing on children's experiences of being diagnosed with epilepsy. This is in contrast to the greater amount of qualitative research conducted on parental views of the diagnosis. The model presented in this research is derived from mainly adult and older adult experiences (both as patients and as relatives of the person with epilepsy). It is not clear to what degree the themes found in this model would apply to children and young people.

It is notable that there are two sets of papers used in this review which share a data set [7 and 20; 6 and 21]. In this systematic review these four papers account for quite an extensive range of conclusions so it is important to recognise the relatively small sample size of participants used (20 participants [7, 20] and 27 participants [6, 21] respectively). This also results in a single, potentially skewed data set, being analysed twice. Given this, there is also a possibility that the super ordinate themes found in this systematic review are similarly affected. While this does not invalidate the conclusions reached in these papers, or the systematic review, the opportunity exists to explore these findings in the wider population.

As noted in the reflexivity section 3.2, the researcher was conducting research on clinicians' experiences of discussing SUDEP at the time of conducting this meta-ethnography. It may be that the results of this research and the researcher's background in psychology influenced the interpretation of this review.

5.3 Conclusion

Despite the extant qualitative research focusing mainly on the experiences of being diagnosed with epilepsy in specific populations, it appears that themes are shared across

populations and experiences and a conceptual model was presented to account for this. Initial symptoms are often spotted by others and diagnosis complications and delays are common. Patients require time to process the diagnosis in terms of informational load and emotional impact. Information is valued more from people (including fellow people with epilepsy or their relatives) rather than leaflets or the internet. Role changes following the diagnosis are usually considered as negative. The relationship with healthcare staff is of central importance not only in relation to the experience of diagnosis, but also as being a source of trust and support.

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Chapter 2: Major Research Project

‘Breaking Good News’: Neurologists’ experiences of discussing SUDEP with patients in Scotland.

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Plain English Summary

SUDEP refers to Sudden Unexpected Death in Epilepsy. It is not known precisely why SUDEP occurs, however people with epilepsy who have a higher number of seizures are at a higher risk of death (although this is a rare event, occurring in only 1/1000 people with epilepsy). Research has shown that American clinicians can be reluctant to discuss SUDEP with their patients in some cases (Miller, Young, Friedman, Buelow & Devinsky, 2014). Research in the UK has also indicated that clinicians may not be regularly informing their patients about the risks of SUDEP. As a result of a Fatal Accident Inquiry (FAI) in 2010, clinicians working in Scotland were advised that they must inform their patients about the risk of SUDEP following a diagnosis of epilepsy except in rare circumstances. This research examined how clinicians have felt discussing SUDEP in this context.

Aim

To understand the experiences of neurologists working in Scotland who have discussed SUDEP with their patients.

Methods

Six Neurologists and four Neurology Registrars who had been working in Scotland for at least one year were recruited. The researcher used a thematic analysis (a type of qualitative analysis) to explore the interviews and identify themes that accounted for their experiences.

Results

Five themes were identified: Clinicians use a 'SUDEP protocol' – this was proposed to be a standardised way of talking about SUDEP; The FAI has diffused into practice through meetings and discussions with colleagues; Clinicians feel ambivalent about discussing SUDEP with patients who are newly diagnosed with epilepsy; Clinicians think that patients will be distressed about hearing about SUDEP, however it is very rare for patients to react badly to the news; Clinicians feel the pressure of needing to discuss SUDEP hinders effective communication with their patients.

Conclusion

This research suggested that the FAI has influenced clinicians working in Scotland and the majority now routinely discuss SUDEP with newly diagnosed epilepsy patients. There is concern, however, that the conversations may have an emotional impact on patients, and that the information patients need to know

about SUDEP is not conveyed appropriately because of the impact of the epilepsy diagnosis. Similar to findings in America (Miller et al., 2014), clinicians value a sense of clinical autonomy. Clinicians feel pressure to discuss SUDEP after an epilepsy diagnosis and they are concerned that patients may fail to fully comprehend the implications of this information, or that the information may be anxiety inducing. This research may be useful for future guideline development.

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Word count: 443

Abstract

Since the findings of a Fatal Accident Inquiry (FAI) in 2010, clinicians working in Scotland have been advised to discuss the risk of Sudden Unexpected Death in Epilepsy (SUDEP) with patients immediately or soon after a diagnosis of epilepsy is made. A thematic analysis was used to describe the experiences discussing SUDEP of 10 clinicians (six Consultant Neurologists and four Neurology Registrars) working in Scotland. Five themes were found:

Clinicians employ a ‘SUDEP protocol’, suggesting there is a standardised way of discussing SUDEP with patients and all clinicians routinely discuss SUDEP with newly diagnosed epilepsy patients; The FAI has diffused into practice through meetings and discussions with colleagues; ‘Breaking Good News’ refers to the ambivalence clinicians feel about discussing SUDEP; ‘Falsely anticipating anxiety’ refers to clinicians anticipating a distressed response from patients despite this very rarely occurring; Clinicians suggest that ‘pressure hinders effective communication’ to patients – suggesting that the pressure to discuss SUDEP early after diagnosis may have an emotional impact on patients and affect the amount of information they can take in. Implications for guideline development are discussed.

Highlights

- A qualitative examination of neurologists’ experiences discussing SUDEP
- Clinicians state they are regularly discussing SUDEP with patients who have newly been diagnosed with epilepsy
- Clinicians feel ambivalent about discussing SUDEP despite indicating the experience is not distressing for themselves, or apparently for their patients

Keywords: SUDEP, epilepsy, qualitative, thematic analysis, neurology practice, breaking bad news

1. Introduction

It has been argued that in the 20th century the risk of death due to epilepsy became minimised then denied by the medical community [1]. It was not until 1996 that the term ‘Sudden Unexpected Death in Epilepsy’ (SUDEP) was proposed. SUDEP refers to the death of a patient with epilepsy which appears to occur without a specific reason [2]. Death can occur

with or without the presence of a seizure (but not due to status epilepticus), be witnessed or un-witnessed, and the term excludes deaths that have occurred due to toxicological or anatomical reasons [3]. The mechanism by which SUDEP occurs is not fully understood [4] however patients can take actions to lower their risk. Chiefly amongst these, people are encouraged to ensure their seizures are well controlled (i.e. occurring at a minimal rate) by adherence to anti-epileptic medication [5, 6]. Other risk factors for SUDEP include: having a greater yearly frequency of generalised tonic-clonic seizures, higher rates of nocturnal seizures [7], and an early age of onset (a higher SUDEP risk occurs in those diagnosed before the age of 16 [8]). SUDEP is a rare event for people with epilepsy, with an incident rate of around 1/1000 [9].

NICE guidelines specify that following a first seizure, patients should see a specialist in the management of epilepsy [10]. Discussions regarding SUDEP with patients should contain 'tailored information' that 'takes account of the small but definite risk of SUDEP' [10, Section 1.3.13, p16] however, access to this information should depend on the certainty of the diagnosis [10]. The American Epilepsy Society and the Institute of Medicine recommend that the increased risk of death associated with epilepsy be disclosed to patients [11, 12].

Harden, Tonberg, Chin, McLellan, and Duncan [13] interviewed adults (aged 18-29) and found they were keen to have a SUDEP discussion with their clinician preferably during the session they are diagnosed with epilepsy, or soon thereafter. Nair, Jack, Meaney, and Ronen [14] conducted focus groups with parents of children with epilepsy and found that they also wished to be informed about SUDEP during their first discussion of epilepsy. Additionally, Nair et al. [14] noted that parents were 'emphatic' that they should not first learn about SUDEP from the internet or an information leaflet. Bereaved relatives of patients who have died from SUDEP are keen that SUDEP risk is discussed with patients and their relatives at

the time of diagnosis, however, they noted that this should be done on a case by case basis with an understanding that the news may have potentially negative consequences [15].

Despite the wishes of patients and the guideline suggestions, the literature has identified that clinicians are not regularly having SUDEP discussions with their patients. Morton, Richardson and Duncan [16] analysed 387 questionnaires when surveying the practice habits of UK based neurologists. Around 70% discussed SUDEP with 'very few' or 'none' of their patients. Similarly, of 1200 American and Canadian neurologists surveyed, less than 7% reported they were routinely discussing SUDEP with all patients [18]. Miller, Young, Friedman, Buelow, and Devinsky [19] used a qualitative approach to understand the practice of American clinicians (Epileptologists, Neurologists, and Advanced Practice Nurses) when discussing SUDEP. A theme of 'moral accountability' was present when clinicians expressed a reluctance to discuss SUDEP with their patients if they felt it was 'morally wrong to give information about a complication that is poorly understood and difficult to prevent' [19, p40]. Clinicians wanted to wait until rapport was built with their patients before discussing SUDEP and there was a reluctance to discuss SUDEP if all treatment options had been tried.

Friedman et al. [18] reported that a perceived negative reaction to discussions of SUDEP were common in their US and Canadian sample of Neurologists. This reaction is perhaps understandable given the findings of research on breaking bad news (BBN). Clinicians may fear a negative response from patients during BBN experiences, and clinicians can feel responsible for the bad news [20, 21]. BBN is especially difficult when there are limited options for treatment [22] or if there is a feeling of inadequacy treating an uncontrollable disease [23]. It is interesting to note that the studies which have examined patients' responses to hearing about SUDEP have not indicated they experience an unduly negative

reaction [13, 24]. The young adults interviewed in Scotland wanted to hear about the risk of SUDEP around the time of diagnosis, and the knowledge that SUDEP may occur led to short lived anxiety which did not appear to influence their health behaviours. The researchers suggested that the patients they interviewed wanted to find out about SUDEP primarily because they had a right to know about this information - not because they could potentially use the information to lower their SUDEP risk [24].

Scottish guidelines issued in 2005 suggested that information about SUDEP was to be considered 'essential' to provide to patients upon diagnosis of epilepsy [25]. Moreover, Neurologists who work in Scotland are likely to be aware of a Sheriff-led Fatal Accident Inquiry (FAI) into the deaths of Erin Casey and Christina Fiorre Ilia in 2010 [26]. This established that two Scottish Health Boards were at fault for not informing these patients, and their parents, of the risk of SUDEP. Following an FAI, the Sheriff will make recommendations as to how to prevent a similar situation occurring in the future. The Scottish Government website states that 'the responsibility for learning any lessons which come out of the inquiry, and for implementing any recommendations made, lie with those who have responsibility for managing the systems in question' [44]. One recommendation was the 'vast majority' of patients should be informed about SUDEP upon being diagnosed with epilepsy or it should be recorded as to why this did not take place. Health Boards can therefore expect a SUDEP discussion to be held with the vast majority of patients following an initial diagnosis. The impact of the medical guideline developments and the FAI on the practice of discussing SUDEP is not yet clear. Research in 2006 suggested that clinicians in the UK were not regularly discussing SUDEP with their patients and this was after the SIGN guidelines had been updated in 2005 [16]. In 2013, Waddell, McColl, and Turner [17], in a retrospective analysis of patients who attended a specialist epilepsy clinic in Scotland, found that a documented discussion of SUDEP occurred in only 4% of the 345 case notes

examined. This may suggest that the FAI in 2010 had a limited impact on the practice of discussing SUDEP for clinicians working in Scotland. It is important to note, however, that the researchers included chronic epilepsy patients in their study and they may not have found a high rate of SUDEP discussions as the Sheriff's recommendation to discuss SUDEP pertained to newly diagnosed patients only. Research in 2015 with young adults in Scotland who had been newly diagnosed with epilepsy indicated they had uniformly been informed about SUDEP after diagnosis [13, 24].

Interestingly, Scottish guidelines were updated again in 2015 and the message regarding the need to discuss SUDEP has changed [27]. The guidelines note that there is not enough evidence to suggest that informing patients about the risk of SUDEP will improve their adherence to medication and the guidelines advise that: 'Counselling about the risks of sudden unexpected death in epilepsy should be considered for patients with epilepsy at an appropriate time for the patient and by an appropriate healthcare professional' [27, p55]. This is clearly less imperative than the former guidelines issued in 2005. The updated guidance may be a recognition that a SUDEP discussion following a diagnosis of epilepsy may not always be necessary or useful.

There is an understanding that clinicians in Scotland have been in a unique position where an FAI has recommended them to have SUDEP conversations with patients following diagnosis. It is not clear as to how Neurologists have interpreted and understood these recommendations and the obligation they feel to practice in this manner. This research was interested in knowing how this has affected their practice and experience of SUDEP discussions. This study was interested in adding to the literature which has assessed if SUDEP discussions are routinely taking place. Given the recent findings that patients do not appear to be overtly distressed learning about SUDEP [13, 24], this study was interested

in understanding clinicians' perceptions of their patients' feelings and if they felt the information about SUDEP would encourage medication adherence. Potential methods of support to facilitate SUDEP discussions between Neurologist and patient were also explored. This research may provide a useful comparative model for countries, health boards or organisations that are considering guideline recommendations for how and when SUDEP is discussed with patients.

1.2 Aim

To explore the experiences of Neurologists when discussing SUDEP with their patients and develop themes to account for these.

The objectives of the research were to understand how the participants discussed SUDEP; how the participants felt when discussing SUDEP (including their thoughts on the impact of the discussion on patients); how they classified good and bad experiences when discussing SUDEP; methods of support utilised or envisioned; feelings about the legal/legislative context to discuss SUDEP and reflections on the practise; as well as assessing if similar themes identified in previous research were present [19].

2. Material and Methods

2.1 Participants

The views of clinicians with a range of experience were sought, therefore Consultant Neurologists and Registrar Doctors were considered for inclusion in the research (typically, Registrar Doctors who specialise in Neurology will have at least 4 years of clinical training in neurology before becoming a consultant). Given the recommendations that 6-12 participants are sufficient for understanding common perceptions and experiences among a

group of relatively homogenous individuals [28], a minimum participant sample size was set at six participants.

2.2 Recruitment and Interview Procedures

The researcher attended a departmental educational meeting of the West of Scotland Neurology Service (a group of Consultant Neurologists and Registrar Doctors) in November 2015 to present information about the research and recruit participants directly. Potential participants were asked to sign up to the research if they were interested and an email was sent to the group's list serve following the meeting seeking interested participants. A set of questions were devised to help guide discussions with participants (Appendix A). These questions were developed by examining the previous literature and by consulting with the Consultant Neurologist Field Supervisor. Supplemental questions were asked based on the content of the interviews. The interview was trialled with the Consultant Neurologist Field Supervisor before conducting interviews with the participants.

Participants were contacted to arrange interview times and the researcher then conducted face to face interviews with participants at their place of work (with one interview being conducted by phone). Participants were given a Participant Information Sheet (Appendix B) and informed consent was obtained before commencing the interview.

2.3 Qualitative Design and Research Procedures

A thematic analysis explored the experiences of clinicians when discussing SUDEP with patients. This particular method was chosen as previous research had used a similar approach to investigate the experiences of American clinicians' practice of discussing SUDEP [19] and it was felt a comparison of practice would be useful. Additionally, given the focus of the research was in describing experiences and practice of the population

studied, a thematic analysis was judged to be more appropriate rather than an analysis which could provide a theory to explain an experience (e.g. grounded theory) or an analysis which is focused more on accounting for a particular individual's experience (e.g. Interpretative Phenomenological Analysis) [30]. The researcher used an inductive approach to analysis [29] as a pre-existing coding frame was not used and themes were constructed from the data, rather than from pre-existing theory. Themes were primarily identified using a semantic approach; taking themes from the explicit statements of participants.

The research procedure was conducted in line with Braun and Clarke's [29] proposed six phases of thematic analysis. This six stages were as followed: 1) After conducting each interview, the interviews were transcribed then read and checked with the audio recording for accuracy. Through this process the researcher noted down initial themes and ideas and became familiar with the data. 2) The data was then coded line by line (see Appendix C for a sample of coded transcript). Codes accounted for 'implicit concerns as well as explicit statements' [30, p50]. Codes were also generated for interesting features of the data (i.e. a code representing a group of other codes, or some broader aspect of the data). It was an aim that the generated codes 'evoke the data' [31]. It was possible, and quite common, for a line of text to have more than one code assigned to it. During this stage, themes of interest began to gradually form and the researcher kept a note of themes that could be explored with subsequent and future transcripts. The research supervisor (a Clinical Psychologist) also coded and commented on an interview data set. 3) As coding progressed, codes were more readily synthesised into themes that were shared between clinicians or accounted for an aspect of the experience that explained a 'patterned response or meaning within the data set' [29, p82]. Therefore, initial transcripts tended to have more codes assigned, whereas later transcripts were more likely to be repeating, or adding to existing themes. A list of all themes was collated and these were clustered into overall themes that could explain the themes found

(Appendix D shows the document that was used to track, alter and arrange these emerging themes). 4) The individual and overall themes were checked with the codes of transcripts, and with the data set as a whole to check for consistency or discrepancy. 5) As this process of checking progressed, overall themes were defined to best account for the data and offer a narrative that could explain the data set. Often this involved altering the composition of overall themes by adding or subtracting individual themes accordingly. 6) The final analysis was expressed with quotes from participants being selected to illustrate the themes found. This offered a final opportunity to check the coherence and explanatory power of the analysis.

2.4 Data Saturation

After the 8th interview no novel themes were found indicating that data saturation had occurred.

2.5 Reflexivity

Blumer [32] describes the assumptions and prior knowledge of a researcher as ‘sensitizing concepts’. Therefore, the researcher’s background as a Trainee Clinical Psychologist is relevant. Issues of support and the psychological impact of breaking bad news were areas of interest to the researcher. The researcher’s own thoughts and interpretations were monitored by keeping a reflective log throughout the research process. Rather than considering the researcher’s opinions as hindering the research, it is acknowledged that these are fundamental to the process and in deriving themes. Of particular relevance may have been the focus on anticipation of distress or anxiety in patients.

2.6 Settings, Equipment and Materials

The researcher used a digital voice recorder to record interviews and data was stored on secure NHS servers.

2.7 Ethical Issues

Data was anonymised and stored in line with the University of Glasgow’s policy on confidential data (http://www.gla.ac.uk/media/media_180727_en.pdf). Anonymity was preserved by redacting location and other identifying information from transcripts. Approval for the study was received from the Research and Development team in NHS Ayrshire and Arran and ethical approval obtained from the school of Medical, Veterinary and Life Sciences at the University of Glasgow. The Research Proposal (Appendix E) for this study was approved by Ayrshire and Arran Research and Development Department and ethical approval granted by the University of Glasgow College of Medical, Veterinary and Life Sciences (Appendix F).

3. Results

Six Consultant Neurologists and four Registrar Doctors participated in the research. The participant details are shown in table 1.

Participant Number	Role	Number of years practising in current role	Length of interview (mins)
Ss 1	Registrar Doctor	5	34.22
Ss 2	Registrar Doctor	4	36.36
Ss 3	Consultant Neurologist	16	32.33
Ss 4	Consultant Neurologist	15	33.11
Ss 5	Registrar Doctor	2	44.07
Ss 6	Registrar Doctor	4	39.54
Ss 7	Consultant Neurologist	10	44.07
Ss 8	Consultant Neurologist	3	35.25
Ss 9	Consultant Neurologist	7	25.20
Ss 10	Consultant Neurologist	2.5	27.24 (Phone interview)

Table 1: Participant characteristics and interview length

Five main themes accounted for the experiences of clinicians when discussing SUDEP: The SUDEP Protocol; Diffusion of the FAI; Breaking Good News – ambivalence discussing SUDEP; Incorrectly Anticipating Distress; and Pressure hinders effective communication. These will be discussed in turn.

3.1 The SUDEP Protocol

Analysis suggested that the practice of informing patients about SUDEP is uniform with only slight variations in practice noted. Clinicians appear to engage in two types of SUDEP conversation; those for patients with newly diagnosed epilepsy (shown diagrammatically in Appendix G) and those for chronic, uncontrolled epilepsy patients.

For chronic uncontrolled epilepsy patients, the SUDEP conversation was invariably used as a means of emphasising the risks associated with poor epilepsy control and in an attempt to encourage medication adherence:

‘so as I said the two patient groups - patients who have chronic epilepsy you are seeing back and they have poor control and you talk about it.’ (Registrar, Ss2)

Sometimes this conversation would occur with people with long standing epilepsy because they had never been made aware of SUDEP:

‘well there are many patients with long standing epilepsy who may have been diagnosed in the days when SUDEP wasn’t discussed but if a long-standing patient were to bring up concern about their risks of seizures or potential of harm then we would have a discussion about that as well.’ (Consultant, Ss3)

Unlike chronic epilepsy patients, clinicians appear to regularly discuss SUDEP with newly diagnosed patients:

'in the last couple of years or the last few months when I was doing first seizure clinics perhaps - almost every new patient, yeah.' (Registrar, Ss6)

There was one very common exception to this rule - the topic of SUDEP is often not mentioned if the patient appears distressed or anxious about the epilepsy diagnosis. Clinicians noted that they will make notes to discuss SUDEP at the next appointment or they rely on Epilepsy Nurses to discuss SUDEP:

'and if they are very anxious during the first consultation I usually do not tell them regarding the diagnosis – regarding the SUDEP risk.' (Consultant, Ss8)

'I heavily rely on my follow-up appointment with nurses where they discuss it far better than me I think, I believe.' (Registrar, Ss5)

Clinicians usually raise the issue of SUDEP first. It was rarer for a patient to initiate a conversation about SUDEP following diagnosis:

Interviewer: 'has there been cases where they know about SUDEP before you raise that?'

'No.' (Consultant, Ss4)

'I've certainly seen some patients who have asked about it. Um, you know I have had, you do get informed patients.' (Registrar, Ss1)

'Um, well at new diagnosis, that's uncommon.' (Consultant, Ss3)

Clinicians often raise the topic of SUDEP towards the end of the diagnosis appointment:

'...towards the end, generally. It's usually the last thing we talk about.' (Consultant, Ss4)

'Not right at the end. I don't want it to be the last thing that they're remembering. I sort of put it in, you know if I've got five things to say it as number three or four.' (Consultant, Ss9)

There is often a 'script' employed which contains the same information and similar phrasing:

'It's probably a personal script I'm not sure I don't know if everybody does that, yes.' (Registrar, Ss6)

'It's the same sentence I use most of the time.' (Consultant, Ss8)

The information tended to include risk factors for SUDEP and it was common to emphasise that the risk of SUDEP is low and modifiable.

'you can explore that a bit further and say yes there is so much risk but it varies and the risk is higher in people who have persistent seizures and it's important that you take your medications and control it and so on.' (Consultant, Ss7)

'[SUDEP is] rare. Uh, that taking appropriate treatment for epilepsy should help avoid it. And I think those are the main things' (Registrar, Ss1)

Many clinicians noted that they will not actually use the term SUDEP, preferring to state there was a risk of harm:

'If I tell the patient he might die because of seizures it may increase anxiety but what I tell is that - in general these are all the potential consequences one can have with epilepsy.' (Consultant, Ss8)

'What I have been doing is discussing that epilepsy can potentially cause serious harm but not quite use the 'death' word straight away.' (Consultant, Ss7)

SUDEP conversations, or the lack of them, were commonly documented to the GP.

'we will either refer you to the nurse specialist who'll cover some of it and if not it will be covered in the next medical clinic. But I'll try and document that as much as possible that it's not done.' (Registrar, Ss6)

Despite the apparent uniformity of practice, clinicians were keen that junior colleagues should develop their own style when discussing SUDEP:

'I'd probably ask them to review the figures and research it themselves as well.'
(Consultant, Ss9)

3.2 Diffusion of the FAI into practice

Clinicians were aware of the FAI and some explicitly noted their thoughts and feelings regarding the ruling having read the inquiry:

'I don't want to point any direction against the sort of judge or the individual people or giving evidence but I don't think that was an amazingly useful event that ruling.'
(Registrar, Ss1)

'it was only after the Sheriff's ruling a few years ago that it kind of became mandatory to bring up SUDEP with patients at the point of diagnosis or soon thereafter.' (Consultant, Ss3)

'I think one thing that probably brought SUDEP to the fore was a fatal accident inquiry.' (Registrar, Ss2)

Others expressed a more vague understanding of the FAI:

'I think there have been rulings, but I wouldn't be able to know the specifics of it, to say that we should all be informing and making [SUDEP] a priority to discuss.'
(Consultant, Ss9)

The suggestion is that the practice of discussing SUDEP has been heavily influenced by the FAI ruling. This was in contrast to the impact of guidelines on SUDEP discussions - Clinicians were aware that guidelines relating to SUDEP practice existed but universally these had not been read.

The FAI appears to have influenced practice by three mechanisms: initial neurology training, teaching days, and discussions with colleagues. Both Registrars and recent Consultants noted that their practice of discussing SUDEP was influenced by their training in neurology:

'[my practice] is from my training days itself- I've not read any guidelines but from the training days itself.' (Consultant, Ss8)

'I think there were a few training sessions that I had attended last time when we were in _____teaching training day.' (Registrar, Ss5)

However, the greater influence on practice appeared to be exerted from colleagues, training and team discussions:

'I've gone to outside meetings and ... those kind of things. And we've had epilepsy training days and we have a monthly training day in epilepsy perhaps comes once a year or things like that.' (Consultant, Ss6)

'Meetings mostly yes - so reasonably formal departmental meetings.' (Consultant, Ss9)

'[I learn it] from peers and epilepsy meetings.' (Consultant, Ss3)

3.3 Breaking Good News – ambivalence discussing SUDEP

Individual clinicians expressed both their support and dissatisfaction with the practice of discussing SUDEP with newly diagnosed patients. This ambivalence extended to the benefits to patients, the feelings regarding the FAI, and whether it is a Breaking Bad News experience or not. In general clinicians stated that SUDEP was an important topic to discuss and patients should be well informed about their condition:

'...obviously you want a well-informed patient, and they're autonomous and should have as much information as you have.' (Registrar, Ss2)

'I think the practice should be that it is important that the patient has all the information of their condition.' (Consultant, Ss7)

Many clinicians viewed SUDEP as a positive topic to discuss as it could increase medication adherence and meant that risk issues could be addressed:

'I think particularly if someone is swithering about compliance for medication – then I think a discussion of SUDEP can make them more adherent to the recommendations.' (Consultant, Ss10)

'I want to frame it and structure it in a way that they think that compliance, lifestyle modification - if I do these two things well from my end then, actually, I am working towards less risk of coming to harm with these seizures. I want to bring a positive approach towards it.' (Registrar, Ss5)

An alternative feeling was also commonly expressed:

'I think there is an argument for should you discuss it should you not and clearly the court has made a decision and that's...but I think there is still an argument as to whether patients should be burdened by this worry' (Consultant, Ss4).

'I think that the guideline that you tell everyone um leaves slightly at the discretion of the clinician. And it's a bit like any other guideline it's a one size does not always fit all. So I think it might be clinically appropriate to leave that to a subsequent consultation to discuss. Particularly if someone is upset having received a diagnosis of epilepsy.' (Consultant, Ss10)

It was this mixture of positive factors, together with the potential anxiety caused to patients, and the perception that the FAI recommendations are mandatory that has created the ambivalence.

Clinicians were unaware of the impact of discussing SUDEP with patients on their behaviour however they hoped that it might influence them positively:

'they probably contribute to good compliance and you know, possibly, they lead to more regular lifestyles and avoidance of binge drinking and other drugs and, I don't, I have no evidence to prove it but that's the hope.' (Consultant, Ss3)

'would hope it would make a difference to them is for them to take control of their condition and try and you know as I say get regular sleep, get the regular meals, avoid alcohol in excess, avoid drugs and take their medication that's what I'm hoping for.' (Registrar, Ss2)

Clinicians were divided as to whether they considered discussing SUDEP a BBN experience:

'no I don't think it's breaking bad news because it's not happened to them it's just, you're just telling them about a potential risk and you've already gone through a lot of potential risks about epilepsy by that stage as well.' (Registrar, Ss2)

'sort of, yeah, it's an educational thing. Sort of breaking bad news.' (Registrar, Ss1)

'I'd say it's breaking good news you know there is this risk but it's usually very, very low.' (Consultant, Ss9)

Clinicians noted that they had not needed or considered personal support for any conversation about SUDEP with patients. Support could come in the form of discussions with colleagues or supervisors however it seemed rare for clinicians to do this.

3.4 Falsely Anticipating Distress

Clinicians stated that they were likely to cause anxiety by discussing SUDEP. There were suggestions that clinicians were unduly worried about what the reaction might be from their patients:

'but I think it's, if it was said to me I think it would be something that would sit you know if I had epilepsy and somebody told me 'you could go to sleep and have a seizure and not wake up' or 'you could have seizure and die from that' I suppose that would worry me, a lot.' (Consultant, Ss4)

'and then very softly say the word death - because it's frightening ... (later on) I think that is more fear in the medics approaching this topic rather than in patients discussing this.' (Registrar, Ss5)

'because if they've not had a seizure for a while and they're on medication then why bring up something that could cause them distress?' (Consultant, Ss3)

Interestingly, there was only one account of a patient reacting badly to the SUDEP discussion. No other clinicians had encountered a bad reaction from any patient when discussing SUDEP. Instead, the general picture is that patients react calmly perhaps only occasionally expressing surprise:

'um so maybe slight surprise - not people getting upset though I don't think in my experience.' (Registrar, Ss2)

'I've not seen any patient giving any anxious reactions so far.' (Consultant, Ss8)

There was some surprise expressed at the fact that patients react so calmly to SUDEP information and some suggestions as to why this is the case. Clinicians felt that the epilepsy diagnosis, or the impact of the condition on their lifestyle, was more distressing than information about SUDEP:

'I think the majority, again, 4 in 5 will respond, pretty surprisingly, without anything. They will just take that as factual information.' (Registrar, Ss6)

'some people are more concerned about their employment, lack of driving so they kind of focus on that.' (Registrar, ss2)

'often the implications of the seizure on their driving activity and other things is of greater concern to them than what is a relatively small risk [of SUDEP].' (Consultant, Ss10)

'rather than [SUDEP] because [lifestyle factors] have the impact you know, do they manage their sleep better, do they take alcohol, are they using recreational drugs on top of that? Are they looking after themselves better? So that's really important.' (Consultant, Ss7)

Clinicians suggested that the cautious approach clinicians take may result in the settled manner patients receive the news. The suggestion was that their way of discussing SUDEP reduced patients' anxieties:

'Uh, I tell in a very smooth manner so that it does not hurt or does not make them very anxious.' (Consultant, Ss8)

'I think to be honest I have never seen it too positive or too negative. There is one standard conversation. And I don't know if maybe I am getting it too easy (laughing) or, if it's just me.' (Registrar, Ss5)

In general clinicians do not find discussing SUDEP with patients an anxiety provoking experience:

'Um...I feel happy talking about it, I feel comfortable talking about it.' (Registrar, Ss2)

There were suggestions that the initial change of practice brought about as a result of the FAI had been anxiety provoking:

'So again when the practice was slowly changing and I thought I should introduce this with every diagnosis. I think initially it was difficult I always felt a little anxious on how to introduce the subject.' (Registrar, Ss6)

It appears that some clinicians were initially anxious about discussing SUDEP however patients' reactions alleviated this anxiety.

'Initially I used to be hesitant but nowadays because it's become routine and after observing the reaction from the patient because it's not an anxiety reaction it is a – they feel it is something like expected.' (Consultant, Ss8)

3.5 Pressure hinders effective communication

Despite reporting that SUDEP is not a distressing experience for them, clinicians did not report universal satisfaction with the requirement to discuss it. It appears that pressure affects clinicians' ability to communicate SUDEP information in a number of ways. In a practical sense, many clinicians note that the main difficulty they had with SUDEP conversations was the limited time they had to discuss the information in addition to diagnosing epilepsy:

'so there's lots of things we have to talk about, or we feel pressure to talk about. We have to talk about lots of different bits that's just one of the other things we have to talk about.' (Registrar, Ss2)

'there is a lot to cover in a 30 minute consultation. You take the history from the patient, sometimes from a witness of a possible event when they've lost consciousness. You're asking them about their past medical history, you're clinically examining them, you are going over investigations they may have had, if they've had imaging, you then sort of discuss the diagnosis you talk about drugs – you don't have, it's something that is kind of shoehorned in – you don't have, it's one of a list of things you need to do.' (Consultant, Ss10)

As a result of the limited time, clinicians wondered if this led to patients being 'overloaded' with information. Clinicians wondered if this affected patients' ability to understand their discussions about SUDEP. This may also be the reason that patients accept the diagnosis so calmly:

'so by the time you come to it the patient already has a lot to absorb. And that's why I think they're already in their minds you know, trying to grasp as much information as they can, so they don't immediately show a response that I have seen.' (Registrar, Ss5)

'I think the new patients are sort of slightly numbed by the time you start to talk about it or are slightly overwhelmed already so they are less likely to engage and ask lots of questions about it I think, in my experience.' (Registrar, Ss2)

There was also a suggestion that the pressure clinicians are under in the initial session to correctly diagnose epilepsy may mean that SUDEP is not given the appropriate emphasis:

'So you've got a very limited time to try and sort of, we say getting the diagnosis right and conveying that to the patient is the primary aim.' (Consultant, Ss7)

In a broader sense clinicians noted their feelings regarding the pressure to discuss SUDEP as a result of the FAI. Clinicians acknowledged the pressure they felt and questioned if the legal system should recommend medical advice to patients:

'and from my perspective too I also need to play safe from the medical legal point of view.' (Consultant, Ss8)

'but whether one feels [discussing SUDEP] is appropriate or not, the fact is if you choose not to do it you're laying yourself open to risk - medical legal risk.' (Consultant, Ss3)

'I think it's probably a bit unfortunate that the way we practice medicine, and this as an example, is not - the decision does not come from the doctors.' (Consultant, Ss4)

'now I think the drive is very much for us to talk about it, bring it up, and also bring it up early talk about it on day 1.' (Registrar, Ss2)

4. Discussion

This research found five main themes that accounted for clinicians' experiences when discussing SUDEP. Clinicians appear to employ a standardised way of discussing SUDEP ('the SUDEP protocol'); the recommendations of the FAI have diffused into their practice through meetings and training; clinicians feel that discussing SUDEP has both negative and positive aspects to it; they report that patients are not distressed by SUDEP information, although they appear to be concerned that it will be distressing for them; and there is concern that the pressure to discuss SUDEP information soon after diagnosis may hinder communication. The results of this study would corroborate recent research which suggests that clinicians in Scotland are regularly discussing SUDEP with newly diagnosed epilepsy patients [13, 24]. The participants in the current study also suggested that SUDEP was not routinely discussed with historically diagnosed epilepsy patients. Both of these findings would not be surprising given that the epilepsy guidelines and the FAI refer to newly diagnosed patients only and have not explicitly stated that the risk of SUDEP should be raised with chronic patients [27].

It was clear that the FAI has had an impact on clinicians practice in Scotland. Even those who were unaware of the specific details of the FAI had their practice impacted. The FAI has created a pressure to discuss SUDEP that is exerted through meetings and discussions amongst Neurologists. Interestingly, this did not seem to be the case for the medical guidelines as clinicians appeared unaware of their content or implications. Systematic reviews suggest that it is difficult to predict if medical guidelines will have an impact on practice [33], however, they are more likely to be successful when introduced alongside rigorous evaluations of their impact [34]. Given that medical guidelines are almost ubiquitous and FAIs into medical practice somewhat rarer, it may be harder to generalise their impact on medical practice. The current study would suggest that compared to medical

guidelines, an FAI has a much greater ability to influence medical practice. This may have been because some clinicians interviewed felt that they were at medical-legal risk if they did not discuss SUDEP.

The influence of the FAI recommendation appears to have resulted in a ‘protocol’ for discussing SUDEP. Clinicians noted that many of the epilepsy diagnosis sessions can take a similar format and SUDEP has clearly been added to the list of necessary topics to discuss. Moreover, there appears to be a standardised way of discussing SUDEP towards the end of diagnosis sessions and a similar approach noted in terms of discussing a ‘risk of harm’, rather than death, and noting measures that patients can take to reduce risk. This SUDEP protocol for chronic epilepsy patients is not as detailed as the protocol for initial diagnosis, however, clinicians appeared to be more effusive towards the benefits of such a discussion.

While the FAI has affected practice, it has also been met with some resistance from clinicians. Miller et al. [19] had found that clinicians in their study suggested it was ‘morally wrong’ to give information about SUDEP because it was poorly understood, and difficult to prevent. Clinicians in the current study were more likely to suggest that the information could have the potential to make a patient anxious or distressed – and this was an unnecessary reaction given the risk of SUDEP occurring. Additionally, the resistance to discuss SUDEP arose because the communicated message was deemed to be less effective following the emotional impact of the epilepsy diagnosis. Psychological research would broadly confirm the clinicians’ sentiments. Anxiety and memory recall for medical information is proposed to be negatively and inversely related (too low, or high a level of anxiety will impair recall [35]). Disclosure of the epilepsy diagnosis may well result in attentional narrowing [36] resulting in less attentional resources available to process and recall SUDEP information [37].

The FAI has created a time pressure for clinicians to discuss SUDEP promptly upon diagnosis and this, again, creates a concern that the information about SUDEP will not be conveyed sympathetically or that patients will not be able to explore or understand the information in sufficient detail. There is, in effect, a potential emotional and informational cost when discussing SUDEP at diagnosis. The patients in the Tonberg et al. [24] study also suggested that SUDEP information should be given in the session following diagnosis, given the emotional impact of receiving an epilepsy diagnosis. Again, this would offer further support, from the patient's perspective, that memory recall is adversely affected in high levels of distress and anxiety [35].

Friedman et al. [18] had found that many clinicians had feared an anxious response in their patients when discussing SUDEP. This feeling was shared by the clinicians in the current study. However, a number of additional statements can be made about this finding. Firstly, although clinicians in the current study were mindful of not creating an anxious response in *patients*, it was clear that they were not anxious about having the discussion themselves. Although some clinicians reported an initial anxiety, most now reported to feel comfortable having the conversation and conveying SUDEP information to patients. This is in concordance with other research which suggests that despite the difficulties involved, doctors in general feel comfortable with BBN experiences [20]. Secondly, despite anticipating an anxious response from patients, there were almost no accounts of this occurring. This finding is corroborated by Harden et al. [13] and Tonberg et al. [24] as patients in these studies reported that SUDEP information created only short-lived anxiety. There may be a suggestion that the cautious approach employed by clinicians, and their anticipation of anxiety, may create the circumstances and atmosphere which results in the calm response from patients. Patients' remember less information when their clinician

appears anxious suggesting that a calm demeanor may facilitate memory recall and lessen emotional impact of a diagnosis [38].

Similar to findings in America, clinicians felt that patients had a right to know about the risk of SUDEP [19]. Clinicians also felt that the conversations about SUDEP might possibly influence adherence to medication. Medication adherence is a multi-factorial process with numerous studies investigating the impact of providing information, self-monitoring, psychological therapy and many more interventions [39]. Evidence would suggest that a conversation with a clinician would, at best, have only a modest influence on rates of medication adherence [40]. Moreover, research has suggested that providing risk information at one point only is not effective in increasing medication adherence in patients with Coronary Heart Disease [41]. Interestingly, research suggests that patients themselves do not feel information about SUDEP impacts their health behaviours however they feel they have a right to all the information about their condition [13, 24].

The Necessity-Concerns Framework (NCF [42]) suggests that for medication adherence to be successful a patient needs to understand the necessity to take their medication and their concerns of possible medication side effects also needs to be addressed. The clinicians in the current study were very clear with regards to the necessity of medication adherence with regards to SUDEP prevention – it may be that exploring the potential side effects of epilepsy medication could also increase adherence rates.

4.1 Limitations

It should be noted that the participants in this study were volunteers and likely to be interested or engaged in the practice surrounding SUDEP. This could have resulted in a more extreme view being expressed either in favour of or against the current practice.

Moreover, clinicians in this study expressed concern that they would be at legal risk if they did not discuss SUDEP with newly diagnosed patients. It would be understandable if those clinicians who do not practice in the legally suggested way may have avoided participating in the current study.

Clinicians did raise additional themes about the practice of discussing SUDEP when asked if they had additional thoughts at the end of the interview. These themes were explored in subsequent interviews however one theme was raised that patients with chronic epilepsy may become more anxious as a result of learning about SUDEP, especially if their epilepsy was not controlled at present. Due to this theme being expressed by a participant late in the recruitment process, this was not able to be fully verified and explored with other data sources. This may have proved interesting to analyse in further detail.

In a similar regard, clinicians expressed that support was available to them however this appeared to be rarely used. Exploring how clinicians utilise and request support from colleagues may be best met with an alternative methodology. Notably, it was felt that clinicians often appeared hesitant and in some cases avoidant when asked about how they utilise or require support. An Interpretive Phenomological Analysis may be more suited to explore this hesitancy and to analyse how the participant is making sense of this phenomenon [43].

4.2 Implications for Research and Practice

Concerns about the impact of SUDEP information on patients remain. Clinicians in the current study expressed interest in establishing whether their conversations about SUDEP made a difference to patients' behaviours. Although qualitative research has suggested that patients profess this is not the case [13, 24], and previous research has suggested that a

clinical conversation in isolation is unlikely to alter an individual's health behaviours [40], it may be useful to quantifiably measure the behaviour change that results from SUDEP conversations.

This research highlighted the impact of guidelines on medical practice. While clinicians were aware that guidelines exist, many were unaware of the contents. It appears that, in this case, FAI recommendations have had a much greater impact on practice than the guidelines suggested by the medical profession itself.

In terms of guideline development, this research has highlighted the additional pressure and concerns that can arise as a result of a 'blanket' approach to practice. This research has strengthened the findings that clinicians value a sense of clinical autonomy [19]. With regards to the practice of discussing SUDEP, removal of this autonomy resulted in clinicians becoming concerned about the quality and emotional impact of the conversation.

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Divide your article into clearly defined and numbered sections. Subsections should be numbered 1.1 (then 1.1.1, 1.1.2, ...), 1.2, etc. (the abstract is not included in section numbering). Use this numbering also for internal cross-referencing: do not just refer to 'the text'. Any subsection may be given a brief heading. Each heading should appear on its own separate line.

Introduction

State the objectives of the work and provide an adequate background, avoiding a detailed literature survey or a summary of the results.

Material and methods

Provide sufficient detail to allow the work to be reproduced. Methods already published should be indicated by a reference: only relevant modifications should be described.

Results

Results should be clear and concise.

Discussion

The **Discussion** section should explore the significance of the results of the work, not repeat them. **Results** and **Discussion** should be separate and may be organized into subheadings. Avoid extensive citations and discussion of published literature.

Conclusions

The main conclusions of the study may be presented in a short Conclusions section, which may stand alone or form a subsection of a Discussion or Results and Discussion section.

Essential title page information

- **Title.** Concise and informative. Titles are often used in information-retrieval systems. Avoid abbreviations and formulae where possible.

- **Author names and affiliations.** Please clearly indicate the given name(s) and family name(s) of each author and check that all names are accurately spelled. Present the authors' affiliation addresses (where the actual work was done) below the names. Indicate all affiliations with a lowercase superscript letter immediately after the author's name and in front of the appropriate address. Provide the full postal address of each affiliation, including the country name and, if available, the e-mail address of each author.

- **Corresponding author.** Clearly indicate who will handle correspondence at all stages of refereeing and publication, also post-publication. **Ensure that the e-mail address is given and that contact details are kept up to date by the corresponding author.**

- **Present/permanent address.** If an author has moved since the work described in the article was done, or was visiting at the time, a 'Present address' (or 'Permanent address') may be indicated as a footnote to that author's name. The address at which the author actually did the work must be retained as the main, affiliation address. Superscript Arabic numerals are used for such footnotes. Please note that proprietary names for drugs should *not* be used in the article title.

Abstract

A concise and factual abstract is required. The abstract should state briefly the purpose of the research, the principal results and major conclusions. An abstract is often presented separately from the article, so it must be able to stand alone. For this reason, References should be avoided, but if essential, then cite the author(s) and year(s). Also, non-standard or uncommon abbreviations should be avoided, but if essential they must be defined at their first mention in the abstract itself.

Highlights

Highlights are a short collection of bullet points that convey the core findings of the article. Highlights are optional and should be submitted in a separate editable file in the online submission system. Please use 'Highlights' in the file name and include 3 to 5 bullet points (maximum 85 characters, including spaces, per bullet point). Highlights are mandatory for Original Reports and Reviews only. They are optional but encouraged for all other article types.

Keywords

Immediately after the abstract, provide a maximum of 6 keywords, using American spelling and avoiding general and plural terms and multiple concepts (avoid, for example, 'and', 'of'). Be sparing with abbreviations: only abbreviations firmly established in the field may be eligible. These keywords will be used for indexing purposes.

Abbreviations

Define abbreviations that are not standard in this field in a footnote to be placed on the first page of the article. Such abbreviations that are unavoidable in the abstract must be defined at their first mention there, as well as in the footnote. Ensure consistency of abbreviations throughout the article.

Acknowledgements

Collate acknowledgements in a separate section at the end of the article before the references and do not, therefore, include them on the title page, as a footnote to the

title or otherwise. List here those individuals who provided help during the research (e.g., providing language help, writing assistance or proof reading the article, etc.).

Formatting of funding sources

List funding sources in this standard way to facilitate compliance to funder's requirements:

Funding: This work was supported by the National Institutes of Health [grant numbers xxxx, yyyy];

the Bill & Melinda Gates Foundation, Seattle, WA [grant number zzzz]; and the United States Institutes of Peace [grant number aaaa]. It is not necessary to include detailed descriptions on the program or type of grants and awards. When funding is from a block grant or other resources available to a university, college, or other research institution, submit the name of the institute or organization that provided the funding. If no funding has been provided for the research, please include the following sentence:

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

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Follow internationally accepted rules and conventions: use the international system of units (SI). If other units are mentioned, please give their equivalent in SI.

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Please submit math equations as editable text and not as images. Present simple formulae in line with normal text where possible and use the solidus (/) instead of a horizontal line for small fractional terms, e.g., X/Y. In principle, variables are to be presented in italics. Powers of e are often more conveniently denoted by exp. Number consecutively any equations that have to be displayed separately from the text (if referred to explicitly in the text).

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Footnotes should be used sparingly. Number them consecutively throughout the article. Many word processors can build footnotes into the text, and this feature may be used. Otherwise, please indicate the position of footnotes in the text and list the footnotes themselves separately at the end of the article. Do not include footnotes in the Reference list.

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General points

- Make sure you use uniform lettering and sizing of your original artwork.
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References

Citation in text

Please ensure that every reference cited in the text is also present in the reference list (and vice versa). Any references cited in the abstract must be given in full.

Unpublished results and personal communications are not recommended in the reference list, but may be mentioned in the text. If these references are included in the reference list they should follow the standard reference style of the journal and should include a substitution of the publication date with either 'Unpublished results' or 'Personal communication'. Citation of a reference as 'in press' implies that the item has been accepted for publication.

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As a minimum, the full URL should be given and the date when the reference was last accessed. Any further information, if known (DOI, author names, dates, reference to a source publication, etc.), should also be given. Web references can be listed separately (e.g., after the reference list) under a different heading if desired, or can be included in the reference list.

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Text: Indicate references by number(s) in square brackets in line with the text. The actual authors can be referred to, but the reference number(s) must always be given.

List: Number the references (numbers in square brackets) in the list in the order in which they appear in the text.

Examples:

Reference to a journal publication:

[1] Van der Geer J, Hanraads JAJ, Lupton RA. The art of writing a scientific article. *J Sci Commun* 2010;163:51–9.

Reference to a book:

[2] Strunk Jr W, White EB. *The elements of style*. 4th ed. New York: Longman; 2000.

Reference to a chapter in an edited book:

[3] Mettam GR, Adams LB. How to prepare an electronic version of your article. In: Jones BS, Smith RZ, editors. Introduction to the electronic age, New York: E-Publishing Inc; 2009, p. 281–304.

Reference to a website:

[4] Cancer Research UK. Cancer statistics reports for the UK, <http://www.cancerresearchuk.org/aboutcancer/statistics/cancerstatsreport/>; 2003 [accessed 13.03.03].

Note shortened form for last page number. e.g., 51–9, and that for more than 6 authors the first 6

should be listed followed by 'et al.' For further details you are referred to 'Uniform Requirements for Manuscripts submitted to Biomedical Journals' (J Am Med Assoc 1997;277:927–34) (see also [Samples of Formatted References](#)).

Journal abbreviations source

Journal names should be abbreviated according to the [List of Title Word Abbreviations](#).

Submission checklist

The following list will be useful during the final checking of an article prior to sending it to the journal for review. Please consult this Guide for Authors for further details of any item.

Ensure that the following items are present:

One author has been designated as the corresponding author with contact details:

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- Full postal address

All necessary files have been uploaded, and contain:

- Keywords
- All figure captions
- All tables (including title, description, footnotes)

Further considerations

- Manuscript has been 'spell-checked' and 'grammar-checked'
- References are in the correct format for this journal
- All references mentioned in the Reference list are cited in the text, and vice versa
- Permission has been obtained for use of copyrighted material from other sources (including the Internet)

Printed version of figures (if applicable) in color or black-and-white

- Indicate clearly whether or not color or black-and-white in print is required.

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Appendix 2. Search Terms for Systematic Review

MEDLINE and EMBASE:

Epilep*.mp. OR seizure*.mp. OR tonic-clonic.mp. OR (tonic adj clonic).mp. OR exp
epilepsy/ OR exp seizure

AND

diagnos*.mp. OR (patient* adj2 experience*).mp. OR (patient* adj2 perspective*).mp. OR
(patient* adj2 reaction*).mp. OR personal experience.mp.

AND

qualitative research.mp. OR qualitative analysis.mp OR exp qualitative research/ OR
content analysis.mp. OR thematic analysis.mp. OR grounded theory.mp. OR Interpretative
Phenomenological Analysis.mp.

PsychINFO:

epilep* OR seizure* OR "tonic clonic"

AND

diagnos* OR patient* N2 experience* OR patient* N2 perspective* OR patient* N2
reaction* OR personal experience

AND

qualitative research OR qualitative analysis OR content analysis OR Interpretative
Phenomenological Analysis OR thematic analysis OR grounded theory

Web of Science

(epilep* OR seizure* OR "tonic clonic")

AND

TOPIC: (diagnos* OR patient\$ NEAR/2 experience\$ OR patient\$ NEAR/2 perspective\$
OR patient\$ NEAR/2 reaction\$ OR personal experience)

AND

TOPIC: (qualitative research OR qualitative analysis OR content analysis OR
Interpretative Phenomenological Analysis OR thematic analysis OR grounded theory)

Appendix 3: Quality Assessment of studies using Walsh and Downe [16] checklist

Study	Clear rationale	Contextualised by literature	Method appropriate	Data collection strategy appropriate	Sample/sampling appropriate	Analytic approach appropriate	Context described	Clear audit trail	Data in support of interpretation	Researcher reflexivity	Sensitive to ethics	Relevance & transferability
[23]	Y	Y	Y	Y	Y	Y	Y	Y	Y	N (<u>Y</u>)	<u>Y</u> (N)	Y
[10]	Y	Y	Y	Y	N	Y	Y	Y	Y	N	Y	Y
[21] β	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
[4]	Y	Y	Y	N	Y	N	Y	Y	Y	Y	N	Y
[7] *	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
[20]*	Y	Y	Y	Y	Y	N (<u>Y</u>)	Y	Y	<u>N</u> (Y)	N	<u>Y</u> (N)	Y
[18]	Y	N	Y	Y	Y	Y	N	Y	N	N	Y	Y
[24]	Y	Y	Y	Y	Y	Y	Y	Y	Y	N	<u>Y</u> (N)	Y
[5]	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
[22]	Y	N	Y	Y	Y	N	N	Y	Y	N	N	Y
[9]	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
[19]	Y	Y	N	Y	N	Y	N	N	Y	N	N	Y
[6]β	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y

*Same sample studied, different focus of study

β Same sample studied, same focus of study, alternative conclusions and themes considered. Differing assessment by the co-rater is indicated in brackets. These were discussed and consensus achieved (shown underlined)

Appendix 4: Studies assessed using Dixon-Woods et al., [17] Fatally Flawed Criteria

	Baca [23]	Buelow [10]	McNelis [4]	Miller 2014a [20]	Nair 2013 [18]	Nair 2016 [24]	Risdale [22]	Thomas [19]
Are the aims and objectives of the paper clearly stated?	Y	Y	Y	Y	Y	Y	Y	Y
Is the research design clearly specified and appropriate for the aims and objectives of the research?	Y	Y	Y	Y	Y	Y	Y	Y
Do the researchers provide a clear account of the process by which their findings were produced?	Y	Y	Y	Y	Y	Y	Y	N
Do the researchers display enough data to support their interpretations and conclusions?	Y	Y	Y	Y	N	Y	Y	N
Is the method of analysis appropriate and adequately explicated?	Y	Y	Y	Y	N	Y	Y	N

Critique of the Nair, Jack, Meaney and Ronen [18] and the Thomas et al. [19] papers.

Nair, Jack, Meaney and Ronen [18] referenced only two papers that investigated families affected by SUDEP and one survey of patients' views to contextualise their research. Thomas et al. [19] employed a thematic analysis to determine themes in their participants' interviews followed by an interpretive phenomenological analysis to develop themes. They

did not provide an explanation of why this method was used. Thomas et al. [19] also did not justify the sample size (having approached 25 participants, 14 were interviewed) and it is unclear when the transcription and interviewing phase was conducted. It was felt that both papers did not give adequate details of the participants in terms of providing details about the social and interpersonal contexts of research participants. The decision trail of Thomas et al's. [19] paper was not clear to follow.

Nair et al. [18] did not use any quotes from participants so it was unclear how the data had been used to support their findings. Thomas et al. [19] could also have considered support for participants who had been distressed discussing their experiences.

Both papers did not meet fatally flawed criteria based on Dixon-Woods et al. [17] criteria and were excluded on this basis.

Appendix 5: Illustrative example of third order themes relating to Article themes for Baca et al. [23]

Study (year)	Method of analysis	Participants	Article themes relating to diagnosis	Detail of themes	Discussion based themes	3 rd Order Themes
Baca et al (2015) [23]	Thematic analysis	37 individual interviews with parents of children with epilepsy who had undergone resective epilepsy surgery	Theme 1: Recognition 'something is wrong'.	Onset of seizures is frightening 'parents describe a feeling of 'desperation to get a diagnosis' p825. Parents are unfamiliar with what epilepsy is Having no idea what it is but needing medical treatment.	Intervention to help families 'learn the language' of epilepsy	Initial onset - frightening Initial onset - Parents don't recognise initial seizures
			Theme 2: Searching and finding: 'a journey around the world and very circuitous'	Overwhelming amount of information 'avalanche of information coming at us' p825 Several parents reported feeling doubted by their doctor Personal and family stress experienced throughout the journey – becoming a caretaker or advocate for their child		Time to process – too much information during the diagnosis Relationship with Healthcare staff- Feel doubted by doctors Role change - Parents become an advocate for their child

Appendix A: Interview Schedule

These questions were designed to facilitate the conversation and often evolved to focus more on specific areas depending on the issues and themes encountered in initial interviews.

Most questions were expanded with follow up prompts. Examples of prompts are given in bullet points below.

Demographics:

Can you describe your role to me?

How long have you been practicing in your current role?

In a typical week or month, how many people would you discuss SUDEP with?

Knowledge

What can you tell me about SUDEP?

What factors influence the risk?

- Prompts: What controllable risks exist? What uncontrollable risks exist?

Are there any guidelines that inform you about SUDEP and what to discuss with patients?

- Prompts: What do SIGN/NICE guidelines say in the subject?

Are there any local NHS policies that affect your practice with regards to SUDEP discussions?

- Prompts: Can you tell me about these?

Typically, what do patients know about SUDEP before you speak to them?

- Prompt: What sources of information have you used?

Practice/Intention

"I am now interested in finding about your typical practice or how you would normally wish to discuss SUDEP with a patient"

When would you typically first discuss SUDEP with a patient?

What influences the timing of when you discuss SUDEP?

- Prompt: are there times or situations when you won't discuss SUDEP?

How is the topic of SUDEP usually first raised?

How do you feel when you know you are about to discuss SUDEP with a patient for the first time?

What key messages do you hope that patients take away with them?

- Prompt: Is there anything you do to help get these messages across to patients?

Experience

“I am interested in hearing about some specific experiences you have had discussing SUDEP with patients.”

Can you tell me about the last time you discussed SUDEP with a patient or carer?

- Prompts: Who initiated this discussion?
- What did you tell the patient/carer about SUDEP?
- What did the patient/carer ask about? What information did they already have? Where had they got this information from?
- How did the patient/carer seem to react/cope with the discussion?
- How did you feel about this discussion?
- What did you do to cope with this discussion?
- Looking back, is there anything you think you could have done/said differently?
- Would you say this was typical as to how SUDEP is discussed?

Can you tell me about a SUDEP discussion that particularly stands out as a difficult discussion?

- Prompts: What was it that was difficult about this discussion?
- Who initiated this discussion?
- What did you tell the patient/carer about SUDEP?
- What did the patient/carer ask about? What information did they already have? Where had they got this information from?
- How did the patient/carer seem to react/cope with the discussion?
- How did you feel about this discussion?
- What did you do to cope with this discussion?
- Looking back, is there anything you think you could have done/said differently?
- Would you say this was typical as to how SUDEP is discussed?

Can you tell me about a SUDEP discussion that particularly stands out as a good or positive discussion?

- Prompts: What was it that was positive about this discussion?
- Who initiated this discussion?
- What did you tell the patient/carer about SUDEP?
- What did the patient/carer ask about? What information did they already have? Where had they got this information from?
- How did the patient/carer seem to react/cope with the discussion?
- How did you feel about this discussion?
- What did you do to cope with this discussion?
- Looking back, is there anything you think you could have done/said differently?
- Would you say this was typical as to how SUDEP is discussed?

Reflection/thoughts on the future

“I would like to hear some of your reflections on your experiences and any thoughts about what could support your practice.”

Looking back on your experiences, what could make SUDEP discussions easier for you or the patient?

- In particular, are there any resources or training courses that could help?

Has your way of discussing SUDEP changed over time?

How do your discussions about SUDEP compare with other times you have had to discuss other ‘bad news’ with a patient?

If you had a difficult experience discussing SUDEP what would you do?

Prompts: Is there anyone you would go to for support?

Is there anything else you would do following a difficult experience?

How do your SUDEP discussions make a difference to patients’ behaviour?

What advice would you give to a junior colleague regarding how to have SUDEP conversations and what to say?



Participant Information Sheet

Clinician experiences of educating patients about SUDEP: How to talk about death when required to do so?

Researcher: Tom Nisbet, Trainee Clinical Psychologist, t.nisbet.1@research.gla.ac.uk

Academic supervisor: Dr Sue Turnbull, University of Glasgow, sue.turnbull@gla.ac.uk,

Field supervisors: Dr Sharon Mulhearn, Consultant Clinical Lead Neuropsychology, NHS Ayrshire and Arran; Dr Saif Ravzi, Consultant Neurologist, NHS GG+C

Research undertaken for a Doctorate in Clinical Psychology, Mental Health and Wellbeing, Institute of Health and Wellbeing, University of Glasgow.

You are being invited to take part in a research study. Before you decide 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 others if you wish. Please ask me 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. Thank you for reading this

1. What is the purpose of the study?

The study will aim to understand and explain the experience of Neurologists or Registrar Doctors who talk to their patients about Sudden Unexplained Death in Epilepsy (SUDEP). This will involve a one-to-one interview between the researcher and a Neurologist/Registrar Doctor. An interview should last between 30-60 minutes and the data collection will aim to be complete by April 2016. The interviews will be analysed qualitatively using a thematic analysis approach to look for themes which explain the experiences of clinicians.

2. Why have I been chosen?

You have been chosen because you are a Neurologist that has worked with people diagnosed with Epilepsy in Scotland for at least one year or because you are a Registrar Doctor who has worked in a Neurology placement and the Consultant has passed your details onto me. The research will involve between 6 – 12 Neurologists.

3. Do I have to take part?

It is up to you to decide whether or not to take part. If you do decide to take part, you will be asked to sign a consent form. If you decide to take part, you are still free to withdraw at any time and without giving a reason.'

4. What will happen to me if I take part?

If you chose to take part the researcher will contact you on the contact details you have provided (please feel free to contact the researcher using the details above if you haven't signed up to the project and wish to partake). The researcher will organise a time with you that is suitable to see you at your place of work and conduct a 30-60 minute interview. The interview will be about your experiences of discussing SUDEP with your patients. The interview will be taped on a digital voice recorder and transcribed onto an encrypted university laptop.

5. What are the possible disadvantages and risks of taking part?

There may be an inconvenience to participation as interviews are expected to last up to 60 minutes. To reduce this researcher will travel to the participants' place of work and offer full flexibility in terms of available times for the interview.

As the overall numbers of clinicians working in this area is small there is a risk that individuals could be recognized. To address this, in addition to removing all names transcripts of the interviews will be redacted for any location information which may identify participants.

There may be a risk that the content of the discussions is upsetting for you (e.g. recalling difficult conversations or clinical decisions).

6. What are the possible benefits of taking part?

You will receive no direct benefit from taking part in this study. The information that is collected during this study aims to explore and identify support mechanisms for this area of practice. Exploring this may help you to recognise these support mechanisms in your own practice. Disseminating the findings may help to develop good practice in this area.

7. Will my taking part in this study be kept confidential?

All information, which is collected about you during the course of the research, will be kept strictly confidential. You will be identified by a pseudonym and any information about you will have your name and location details removed so that you cannot be recognised from it. Additionally, because the sample size is small, transcripts of the interviews will be redacted for any location information which may identify participants.

Data will be stored at the University of Glasgow for ten years after the research is completed in line with the University's Code of Good Practice in Research.

Data will be anonymised and stored in line with the University of Glasgow's Code of Good Practice in Research (http://www.gla.ac.uk/media/media_227599_en.pdf). Anonymity will be preserved by recruiting from across Scotland and redacting location and other identifying information from transcripts. An encrypted university laptop will be used to store the transcribed recording. The transcription will be satisfactorily checked for accuracy against the recording, and then the recording will be erased.

8. What will happen to the results of the research study?

The results of the study will be written up to form part of a completed Clinical Psychology Doctoral dissertation. Additionally, the research will be written up with the goal of being published in a medical journal.

9. Who is organising and funding the research?

The research forms part of the Doctoral award conferred by the University of Glasgow funded by NHS Education for Scotland.

10. Who has reviewed the study?

The research has been reviewed and approved by the College of MCLS Ethics committee at the University of Glasgow.

11. Contact for Further Information

Tom Nisbet, Trainee Clinical Psychologist, t.nisbet.1@research.gla.ac.uk.

Academic supervisor: Dr Sue Turnbull, University of Glasgow, sue.turnbull@gla.ac.uk, 0141 2113937

Thank you for taking part in this study

Appendix C: Sample of transcript coding (5th Participant)

	Code	Theme
First of all I try and enquire about your understanding of, you know, what epilepsy is and what risk you have. Normally I lead it with an open question - so what do you know about epilepsy? You know, what is the condition? And normally this leads me to the conversation.	Gain understanding of epilepsy from patient Use open questions	
I normally say that you know there is a risk associated with epilepsy – that people, we know people can come to harm with epilepsy - especially if it is not treated or medication is missed. This is a small risk. But it is there. That is why we have to mention it. Epilepsy is treatable in the majority of cases and if it is well-controlled the risk goes down. That’s why it’s even more important to know there is a risk and that the risk can be reduced further. That’s why I don’t want to try and terrify individuals by saying ‘oh my god – it’s death’ if, you know, the minute that word is mentioned people don’t engage and, you know, they don’t register much after that. So I bring it as a positive thing and say “there is a risk but it is controllable and more defiable if you’re on treatment and if you are compliant with your medication in the majority of the cases that risk can be reduced. But it's worthwhile knowing so that you can maybe change some lifestyle if it is risky and may be involved lots of alcohol, late nights you know, awakenings or lack of sleep and travelling too much and all that so... I kind of don’t directly say ‘SUDEP - this is the risk factor, there are people who die with it’ I introduce it as one of the factors that need to be aware of and how that can be improved to bring a positive - and they register it more and you know, they are less terrified and I think they say “oh well it’s there but we can do something about it”	Risk with sudep but small Untreated increases risk but its small Don’t want to terrify individuals ‘death’ and they don’t engage Compliance means risk is reduced Lifestyle factors affect risk Introduce SUDEP as a positive	Sudep for adherence Emphasises rarity Anticipating anxiety Difficulty with term ‘death’ SUDEP for adherence SUDEP for adherence
So it’s almost like a way of encouraging them to get their epilepsy under control?		
Yeah controlled	SUDEP for control	
You mentioned that you know as soon as the word ‘death’ is mentioned it can be a kind of, you know, not wanting to terrify people - is that a kind of typical reaction you’ve seen before?		
I think have not seen it because I have seen that people when you talking to them – the problem is at the first seizure clinic there is little time and so	Not seen terrified patients before	Patients not upset by SUDEP info

<p>much to discuss. The driving, their lifestyle, diagnosis, prognosis, certainty of diagnosis and everyone has their own questions as to what is my next generation risk and all that - so by the time you come to it the patient already has a lot to absorb. And that's why I think they're already in their minds you know, trying to grasp as much information as they can, so they don't immediately show a response that I have seen. They kind of go like blank face and trying to get what you are saying. So when they come back for the second or third appointment that's when they open up and say "you mentioned about that and you know what's my risk? And I have been thinking about it..." so I do you see that they kind of cling onto that information but not immediately react.</p>	<p>Time factor to discuss many things</p> <p>A lot to absorb for patients</p> <p>Patients talk more about sudep in second or third apt</p> <p>Retain information about SUDEP in first apt?</p>	<p>Time pressure</p> <p>Information overload</p> <p>Information overload</p>
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Appendix D: List of Overall Themes

The SUDEP protocol

SUDEP for initial SUDEP for chronic – ss3 wait for chronic to raise it?

Won't discuss if chance is remote ss9

More SUDEP conversations now than in the past (even for ST3 ss6) ss9

Difficulty with the term – death, ss7 ss8 cf ss8 write down the term ss9

sudep raised by clinician (Rare for patients to be aware of SUDEP cf ss8) ss8

Discuss at diagnosis –cf ss4 cf ss7 staggered approach to SUDEP discussion

Won't discuss if patient is distressed at diagnosis

Don't discuss – patients reaction

Consider written information ss9

Discuss SUDEP at end of session ss8 cf ss9 in the middle

Use of script ss8

Rapport important ss6 ss7

Will document sudep conversation ss8

(Risks for SUDEP - Don't know what causes SUDEP, Nocturnal risk factor, Lifestyle factors)

Epilepsy nurses can discuss sudep in more detail – better, soon ss7 ss8 ss9 ss10

Diffusion of the FAI into practice

Unaware of guidelines cf ss4 (encourage junior colleagues to read guidelines)

Unaware of policy

Desire for clinical judgement (moral autonomy – Miller paper)

Continuing debate regarding SUDEP ss3 ss4

Learning Practice

knowledge of SUDEP through teaching (Reg) (Cons ss8)

knowledge of sudep through colleagues

SUDEP conversations shaped by experience ss6 interesting, ss7

SUDEP has additional 'weight' ss8

Breaking Good News – ambivalence discussing sudep

Discuss because of FAI – good thing ss5 – bad thing ss3 –pressure, bad thing ss7

Uk more litigious and educated so will discuss SUDEP more at diagnosis ss8

Consider important to discuss cf ss4

Sceptical of the FAI ss10

Clinical judgement mentioning risk factors

Hopes of discussing SUDEP:

SUDEP for adherence

SUDEP to share info [patients have a right to know? Help increase adherence from health model – actually need to increase understanding that medication is not harmful]

SUDEP for engagement

View SUDEP as a positive cf ss4 (because anticipating distress??)

Impact of communicating - Unaware of effect of communicating SUDEP ss8

Impact – conversations make a difference ss6 ss7 ss9 (cf health model)

BBN

Ambivalence

Definitely not BBN ss3 ss5

Same ss4 ss7

Definitely – ss9

Falsely Anticipating distress

Patients not upset by SUDEP info cf SS3 – concerned ss4 ss6 react well –this is surprising ss8, possibly because avoid discussing when distressed ss8

Patient not surprised ss7 cf ss3 ss4 ss8 this is surprising itself ss9 cf ss10 - Sudep not surprising given dramatic nature of seizures

Driving more important than SUDEP

SUDEP helpful for chronic

Relatives more worried than patients

no experience of difficult SUDEP conversation cf ss3 ss10

patients react well better if they have read about it ss5 cf ss7

epilepsy diagnosis worse than SUDEP

Rare for patients to ask more about SUDEP ss6 ss8

Chronic uncontrolled patients can become unduly worried about SUDEP ss8

more fear in medics than patients Ss5 ss10 (colleagues activated patients would respond badly to SUDEP)

anticipating anxiety - impact of discussing SUDEP after epilepsy diagnosis

even in chronic patients – unwise to mention ss3

emphasise rarity – minimise risk

gentle delivery

Clinicians reaction

Not distressed discussing SUDEP cf ss4 ss6 – not initially ss6 – interesting, not bothered despite not thinking they should discuss SUDEP ss7 ss8 – possibly because have seen their natural reaction

Uncomfortable answering questions at first

More worry about getting epilepsy controlled than sudep (this is clinicians perceptions, they don't actually know this)

SUDEP not difficult in relation to other conditions – similar to asthma diabetes ss3 cf ss7ss8

Support

Encouraged to share difficult experiences (Reg)

Discuss with colleagues – feedback

Avoidance of acknowledging support/feelings? Ss6 logistics ss7 ss8 ss9 ss9-discuss with family ss10 interesting

Pressure hinders effective communication

Time pressure - Problem is discussing SUDEP at diagnosis ss2 ss6 ss8

Pressure to get diagnosis correct

Patients priority isn't SUDEP - Patients should decide relevance of discussing SUDEP ss6 is clinical practice best influenced by legal system? Ss3 ss4

information overload

Appendix E: Accepted Research Protocol

Cover Page

Name of assessment: MRP Proposal

Title: Clinician experiences of educating patients about SUDEP: How to talk about death when required to do so?

Matriculation Number: 2109093

Date of submission: 23/05/2015

Version number: 8.4

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Abstract

Sudden Unexplained Death in Epilepsy (SUDEP) occurs in roughly 1/1000 patients with epilepsy. Upon diagnosis, UK guidelines specify that patients should be given information about how to lower their risk of SUDEP. Despite this, research demonstrates that clinicians often fail to have SUDEP conversations with their patients. Although numerous factors affect clinicians' experiences of Breaking Bad News (BBN), the research into clinicians' experiences of discussing SUDEP is limited to an American study. Given that the American SUDEP guidelines are not as prescriptive as the Scottish guidelines, their experience is different. This research is interested in understanding the experiences of clinicians in Scotland when discussing SUDEP with their patients. Neurologists will be interviewed about their experiences of discussing SUDEP with patients. Line by line analysis of transcripts will help identify salient themes in the clinicians' experiences. It is hoped this research will identify support for practice.

Introduction

It has been argued that in the 20th century the risk of death due to epilepsy became minimised and then denied, despite the evidence this was not the case (Hanna & Panelli, 2011). It was not until 1996 that the term 'Sudden Unexplained Death in Epilepsy' (SUDEP) was proposed. SUDEP refers to the death of a patient with epilepsy which appears to occur without a specific reason (Nashef, 1997). Death can occur with or without the presence of a seizure (but not due to *status epilepticus*), be witnessed or un-witnessed, and the term excludes deaths that have occurred due to toxicological or anatomical reasons (Scorza, Cysneiros, Albuquerque, Scattolini, & Arida, 2011). Despite the name, people with epilepsy can take actions to reduce their risk of SUDEP. Chiefly amongst these, people are encouraged to ensure their seizures are well controlled (i.e. occurring at a minimal rate) by adherence to anti-epileptic medication (Hughes, 2009; Scorza, Arida, Terra & Cavelheiro, 2010). Other risk factors for SUDEP include: having a greater yearly frequency of generalised tonic-clonic seizures, an early age of onset (the highest SUDEP risk occurs in those diagnosed before the age of 16 (Hesdorffer et al., 2011)), winter temperatures, and medication use (with polytherapy carrying a higher risk (Scorza et al., 2011)). SUDEP is a rare event for people with epilepsy, with an incident rate of around 1/1000 (Gayateri, Morrall, & Jain, 2010).

NICE guidelines specify that following a first seizure, patients should see a specialist in the management of the epilepsies (NICE, 2012). Discussions regarding SUDEP with patients should contain 'tailored information' that 'takes account of the small but definite risk of SUDEP' (NICE, 2012, Section 1.3.13, p16) however, access to this

information should depend on the certainty of the diagnosis (NICE, 2012). The American Epilepsy Society and the Institute of Medicine recommend that the increased risk of death associated with Epilepsy be disclosed to patients (Hirsch et al., 2011; Institute of Medicine, 2012).

Harden, Tonberg, Chin, McLellan, and Duncan (2014) interviewed adults (aged 18-29) and found they were keen to have a SUDEP discussion with their clinician. Nair, Jack, Meaney, and Ronen (2013) conducted focus groups with parents of children with epilepsy and found that they also wished to be informed about SUDEP during their first discussion of epilepsy. Additionally, Nair et al. (2013) noted that parents were 'emphatic' that they should not first learn about SUDEP from the internet or an information leaflet.

Despite the wishes of patients and the guideline suggestions, clinicians are not regularly having SUDEP discussions with their patients. Morton, Richardson and Duncan (2006) analysed 387 questionnaires when surveying the practice habits of UK based neurologists. Around 70% discussed SUDEP with 'very few' or 'none' of their patients. Similarly, of 1200 American and Canadian neurologists surveyed, less than 7% reported they were routinely discussing SUDEP with all patients (Friedman, Donner, Stephens, Wright & Devinsky, 2012).

Friedman et al. (2012) reported that a perceived negative reaction to discussions of SUDEP were common in their US and Canadian sample of Neurologists. This reaction is perhaps understandable given the findings of research on breaking bad news (BBN). Clinicians may fear a negative response from patients during BBN

experiences, and clinicians can feel responsible for the bad news (Schildmann, Cushing, Doyal & Vollmann, 2005; Tesser, Rosen, & Tesser; 1971). BBN is especially difficult when there are limited options for treatment (Ptacek & Eberhardt, 1996), or if there is a feeling of inadequacy treating an uncontrollable disease (DoH, 2003).

Clinicians value a sense of autonomy regarding the disclosure of bad news. In a survey of FY1/FY2 Doctor equivalents, Schildmann et al. (2005) found that although the majority of clinicians believed patients should be aware of life-threatening illnesses, a third felt that Doctors should be able to decide whether to give patients bad news or not. Even experienced clinicians appear to find BBN experiences emotionally difficult (Ptacek, Ptacek & Ellison, 2001) and it is recognised that a lack of emotional support from healthcare colleagues can act as an 'institutional barrier' to good practice (Dosanjh, Barnes, & Bhandari, 2001).

Miller, Young, Friedman, Buelow, and Devinsky (2014) used a qualitative approach to understand the practice of American clinicians (Epileptologists, Neurologists, and Advanced Practice Nurses) when discussing SUDEP. A theme of 'moral accountability' was present when clinicians had some doubt whether to discuss SUDEP with their patients or not. Clinicians wanted to wait until rapport was built with their patients before discussing SUDEP and there was a reluctance to discuss SUDEP if all treatment options had been tried.

Scottish guidelines consider SUDEP to be essential information for patients (SIGN, 2005). Neurologists who work in Scotland are also likely to be aware of a judge-led

fatal accident enquiry into the deaths of Erin Casey and Christina Fiorre Ilia (2011). This established that two Scottish Health Boards were at fault for not informing these patients, and their parents, of the risk of SUDEP. One recommendation was the 'vast majority' of patients should be informed about SUDEP or it should be recorded as to why this did not take place. Health Boards can therefore expect a SUDEP discussion to be held with the vast majority of patients following an initial diagnosis. Waddell, McColl, and Turner (2013), in a retrospective analysis of patients who attended a specialist epilepsy clinic in Scotland, found that a documented discussion of SUDEP occurred in only 4% of the 345 case notes examined.

The proposed study is interested in understanding how the Scottish guidelines and legislative context has affected the experience of Neurologists when discussing SUDEP. This study will also investigate the resources clinicians employ to enable this discussion. This research will help identify potential methods of support to facilitate SUDEP discussions between neurologist and patient. Additionally, this research may provide a useful comparative model for countries, health boards or organisations that are considering their guideline recommendations for how SUDEP is discussed with patients.

Aim

To explore the experiences of Neurologists when discussing SUDEP with their patients and develop themes to account for these.

The initial objectives of the proposed study will be to understand how the participants discuss SUDEP; how they classify good and bad experiences when discussing SUDEP; methods of support utilised or envisioned; feelings about the legal/legislative context to discuss SUDEP; as well as identifying if similar themes identified in previous research are present (Miller et al., 2014).

Methods

Participants

Participants in the study will be practicing Consultant Neurologists with at least one year's experience managing patients with seizures and epilepsy as a Consultant in Scotland. Less experienced Registrar Doctors will also be approached for interviews. Registrar Doctors will be eligible for inclusion if they have worked for any length of time in a neurology placement and have had a minimum of two discussions about SUDEP.

Recruitment Procedures

The Field Supervisor (a Consultant Neurologist) will send details of the proposed

study to the West of Scotland Neurology email circulation list. This list goes to approximately 30 Neurologists working across the West of Scotland through NHS Greater Glasgow and Clyde Health Board. **Registrar Doctors are also subscribed to this list.** This will request that the researcher attend a meeting of the group in September 2015 to present information about the research and recruit participants directly. Participants will be given a participant information sheet, asked to consent to the research and arrange interview times, or provide contact details, at this stage. The researcher will **ask that the Field Supervisor** emails all members of the email circulation list following the lunchtime meeting. The email will give information about the research and ask members to contact the researcher or Field Supervisor if interested in participating. Registrar doctors will be informed that their choice to participate in research will not affect their training and any data would be confidential.

Participants will be contacted to arrange interview times and the researcher will bring a participant information sheet and allow time to answer questions prior to the interview beginning so that informed consent can be obtained. Participants will be asked if they wish to provide contact details so that the researcher can arrange a follow up appointment if required.

Qualitative Design and Research Procedures

A thematic analysis will explore the experiences of clinicians when discussing SUDEP with patients. The analysis will take a contextualist approach focussing primarily on the manner neurologists make sense of their experiences, while

retaining the scope to analyse how broader contextual factors may have influenced their understanding. The analysis will focus on providing a description of the data set as a whole, rather than looking at one aspect of the data in detail. The researcher will use an inductive approach to analysis (Braun & Clarke, 2006) as a pre-existing coding frame will not be used and themes will be constructed from the data itself, rather than from pre-existing theory. Themes will primarily be identified using a semantic approach; taking themes from the explicit statements of participants.

One-to-one interviews between the researcher and the interviewee will be recorded using a digital voice recorder. Interviews will last between 30-60 minutes. A set of questions will help guide discussions. These questions were developed by examining the previous literature described above and with consultation of the Consultant Neurologist Field Supervisor. Supplemental questions may be asked based on the content of the interviews. The interview will be trialed with the Neurologist Field supervisor before conducting interviews with the participants.

The research procedure will be conducted in line with Braun and Clarke's (2006) proposed six phases of thematic analysis. After conducting each interview, the transcriptions will be read and checked with the audio recording for accuracy. Through this process the researcher will note down initial ideas and become familiar with the data. It is proposed that the data is then coded line by line. This will generate codes that account for 'implicit concerns as well as explicit statements' (Charmaz, 2006, p50). Codes may also be generated for any interesting feature of the data (i.e. a code representing a group of other codes, or some broader aspect

of the data). In effect the generated codes should in some way 'evoke the data' (Clarke and Braun, 2013).

After initial coding, a process of focused coding will synthesise codes into salient themes that account for clinicians' experiences. Themes will be developed which represent a 'patterned response or meaning within the data set' (Braun & Clarke, 2006, p82). Ultimately, themes will explain something meaningful to the research question therefore the prevalence of the theme occurring in the data may or may not be important.

Themes will be checked with the codes of transcripts, and with the data set as a whole to check for consistency or discrepancy. As the process continues for each new interview transcription, the themes will become further refined. Themes of interest can be verified and explored with other data sources so that the themes produced represent something meaningful to the data set. Negative cases will also be of interest should the data provide them (Strauss & Corbin, 1990). The final report will include a description of each theme found with examples from the data set.

Data Saturation

Analysis will continue until data saturation is achieved; this will be indicated when novel themes are no longer identified in the data set. This may involve follow up interviews with some clinicians and the possibility of conducting interviews with

Registrar Doctors. It is suggested that at least 6 participants are required at a minimum to evaluate when data saturation may occur.

Reflexivity

Blumer (1954) describes the assumptions and prior knowledge of a grounded theory researcher as 'sensitizing concepts'. Therefore, the researcher's background as a Trainee Clinical Psychologist will be relevant. Issues of support and the psychological impact of breaking bad news have already been mentioned in this proposal as these are potential areas of interest to the researcher. The researcher's own thoughts and interpretations will be monitored by keeping a reflective log throughout the research process. Rather than considering the researcher's opinions as hindering the research, it is acknowledged that these are fundamental to the process and in deriving a theory.

Settings, Equipment and Materials

The researcher will use a digital voice recorder, transcribing equipment and an encrypted laptop. The researcher will travel out to interview clinicians in their NHS work setting. The researcher will ask that there is a quiet room available for 45-60 minutes that is suitable for the use of recording equipment. If it is not possible to arrange a face to face interview the use of Skype™ will be considered.

Financial Issues

Travel to potential sites will be claimed from the NHS employer. It is anticipated that the only costs incurred will be for the photocopying of consent and information forms.

Health and Safety Issues

The researcher will follow the health and safety procedures of the NHS settings where the interviews take place. The interviews will involve discussion of difficult experiences and the researcher will discuss support options available to the clinician if this is felt appropriate. Identifying and exploring such experiences is an aim of the research, however, if the interviewee expresses ongoing psychological distress they will be signposted to relevant services. The clinicians will be informed that they can terminate the interview at any point.

Ethical Issues

Data will be anonymised and stored in line with the University of Glasgow's policy on confidentiality data (http://www.gla.ac.uk/media/media_180727_en.pdf). Anonymity will be preserved by recruiting from across Scotland and redacting location and other identifying information from transcripts. An encrypted university laptop will be used to store the transcribed recording. The transcription will be satisfactorily checked for accuracy against the recording, and then the recording will be erased. Given that no NHS patients will be approached in this study, the researcher will obtain ethical approval for the research through the University of

Glasgow. Approval for the study will be sought from the Research and Development team in Greater Glasgow and Clyde Health Board.

Timetable

A full proposal will be submitted for approval by March 16th 2015. Ethical approval will be sought for the research by the beginning of July 2015. Recruitment will begin in September 2015 and it is proposed that interviews are concluded by December 2015. The analysis will be written by February 2016 with a first draft completed by April 2016.

Practical Applications

This research will help develop testable hypothesis and may indicate why so few clinicians have SUDEP discussions with their patients (Morton et al., 2006; Waddell et al., 2013). Given the legal imperative of these discussions, it is hoped this research will be useful in determining support to aid practice. It is planned that the research be submitted for publication to a medical journal.

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Appendix F: Ethical Approval from University of Glasgow MVLS.

7 September 2015

Dr Susan Turnbull
Mental Health and Wellbeing
Admin Building
Gartnavel Royal Hospital
1055 Great Western Road
Glasgow G12 0XH

Dear Dr Turnbull

MVLS College Ethics Committee

Project Title: Clinician experiences of educating patients about Sudden Unexplained Death in Epilepsy (SUDEP): How to talk about death when required to do so?

Project No: 20015005

The College Ethics Committee has reviewed your application and has agreed that there is no objection on ethical grounds to the proposed study. It is happy therefore to approve the project, subject to the following conditions:

- Project end date: 30 September 2016.
- The data should be held securely for a period of ten years after the completion of the research project, or for longer if specified by the research funder or sponsor, in accordance with the University's Code of Good Practice in Research:
(http://www.gla.ac.uk/media/media_227599_en.pdf)
- The research should be carried out only on the sites, and/or with the groups defined in the application.
- Any proposed changes in the protocol should be submitted for reassessment, except when it is necessary to change the protocol to eliminate hazard to the subjects or where the change involves only the administrative aspects of the project. The Ethics Committee should be informed of any such changes.
- You should submit a short end of study report to the Ethics Committee within 3 months of completion.

Yours sincerely



Professor William Martin
College Ethics Officer

Approval20015005.docx

Appendix G: The SUDEP Protocol diagram for initial diagnosis conversations

