



Casselden, Louisa (2015) An exploration of body image and self-esteem in adolescents with implantable cardioverter defibrillators (ICDs): a qualitative study and clinical research portfolio. D Clin Psy thesis

<http://theses.gla.ac.uk/6700/>

Copyright and moral rights for this thesis are retained by the author

A copy can be downloaded for personal non-commercial research or study, without prior permission or charge

This thesis cannot be reproduced or quoted extensively from without first obtaining permission in writing from the Author

The content must not be changed in any way or sold commercially in any format or medium without the formal permission of the Author

When referring to this work, full bibliographic details including the author, title, awarding institution and date of the thesis must be given



University
of Glasgow

VOLUME I

**An Exploration of Body Image and Self-Esteem in Adolescents with
Implantable Cardioverter Defibrillators (ICDs): A Qualitative
Study and Clinical Research Portfolio**

(Volume II bound separately)

Louisa Casselden, MA (Hons), MSc

Submitted in partial fulfilment of the requirements for the degree of Doctorate in
Clinical Psychology

Institute of Health & Wellbeing, University of Glasgow

College of Medical, Veterinary and Wellbeing

University of Glasgow

September 2015

©Louisa Casselden



Declaration of Originality Form

This form **must** be completed and signed and submitted with all assignments.

Please complete the information below (using BLOCK CAPITALS).

Name LOUISA CASSELDEN
Student Number 2058547
Course Name DOCTORATE IN CLINICAL PSYCHOLOGY
Assignment Number/Name CLINICAL RESEARCH PORTFOLIO

An extract from the University's Statement on Plagiarism is provided overleaf. Please read carefully THEN read and sign the declaration below.

I confirm that this assignment is my own work and that I have:

- Read and understood the guidance on plagiarism in the Doctorate in Clinical Psychology Programme Handbook, including the University of Glasgow Statement on Plagiarism ✓
- Clearly referenced, in both the text and the bibliography or references, **all sources** used in the work ✓
- Fully referenced (including page numbers) and used inverted commas for **all text quoted** from books, journals, web etc. (Please check the section on referencing in the 'Guide to Writing Essays & Reports' appendix of the Graduate School Research Training Programme handbook.) ✓
- Provided the sources for all tables, figures, data etc. that are not my own work ✓
- Not made use of the work of any other student(s) past or present without acknowledgement. This includes any of my own work, that has been previously, or concurrently, submitted for assessment, either at this or any other educational institution, including school (see overleaf at 31.2) ✓
- Not sought or used the services of any professional agencies to produce this work ✓
- In addition, I understand that any false claim in respect of this work will result in disciplinary action in accordance with University regulations ✓

DECLARATION:

I am aware of and understand the University's policy on plagiarism and I certify that this assignment is my own work, except where indicated by referencing, and that I have followed the good academic practices noted above

Signature LOUISA CASSELDEN

Date: 23/9/2015

Acknowledgements

I would like to extend my deepest thanks to the young people who participated in this study. They gave up their time to share their personal stories and experiences with me, so that treatment and services for other young people with similar difficulties might be improved in the future. Thank you. The courage and strength that these young people and their families display in the face of their health conditions is truly inspiring.

I would also like to give my heartfelt thanks to my academic supervisors Dr Alison Jackson and Dr Sarah Wilson. Without their patience and direction I do not think my research would have come together as it has. Thank you for containing my anxiety, your words of wisdom have been invaluable. Thank you also to my field supervisor, Dr Kathleen McHugh, who guided me through the pitfalls of recruitment and data collection, and the cardiac nurses at the Royal Hospital for Sick Children, for their advice. I would also like to acknowledge the National Institute for Cardiovascular Outcomes Research for the statistics they provided regarding prevalence of ICD devices in the UK.

My thanks would not be complete without acknowledging my amazing family and friends who have helped to keep my sanity throughout the last three years. My wonderful husband Chris has been a pillar of strength and encouragement, thank you from the bottom of my heart. Also, I need to thank all the brilliant DClinners, especially Jill, Stephanie and Fran for all the comforting hugs, wine and laughs. It has been a privilege getting to know such a wonderful group of people. Edel, thank you for reading everything and giving me your words of advice and encouragement, I will definitely return the favour when it is your turn. Also, thank you to Rachel, Jessica and Christine. You have all patiently allowed me to let off steam, sympathising and encouraging me even when you have no idea what I was talking about. Finally, thank you to my dear parents Christina and David, who have always believed in me. You are the reason I have come so far. Thank you for everything.

<u>Volume 1- Table of Contents</u>	<u>Page</u>
CHAPTER ONE: SYSTEMATIC LITERATURE REVIEW	1
<u>Psychosocial Adaptation in Parents of Children with Cardiac Arrhythmias: A Systematic Review</u>	
Abstract	2
Introduction	3
Rationale & Objectives	5
Method	6
Results	8
<i>Figure 1:Flow Diagram of Selection Process</i>	10
<i>Table 1: Overview of Studies</i>	11
<i>Table 2:Comparisons of Mothers and Fathers</i>	23
Discussion	24
Conclusions	29
References	30
CHAPTER TWO: MAJOR RESEARCH PROJECT	37
<u>An Exploration of Body Image and Self-Esteem in Adolescents with Implantable Cardioverter Defibrillators (ICDs): A Qualitative Study.</u>	
Plain English Summary	38
Abstract	39
Introduction	40
Aims	43
Method	43
<i>Table 1: Inclusion and Exclusion Criteria</i>	44
<i>Figure 1:Flow Diagram for Recruitment</i>	45
<i>Table 2: Demographic Information of Participants</i>	46
<i>Table 3: Procedure for Data Analysis with IPA</i>	47
Results	49
<i>Table 4: Superordinate and Emergent Themes</i>	50
Discussion	64
<u>Volume 1- Table of Contents</u>	<u>Page</u>

Conclusions	71
References	72
CHAPTER THREE: REFLECTIVE ACCOUNT	79
<u>Formulating Therapeutic Resistance</u>	
Abstract	80
CHAPTER FOUR: REFLECTIVE ACCOUNT	81
<u>Working Across Services: A Reflection on Service Issues</u>	
Abstract	82
APPENDICES	
Systematic Literature Review	
Appendix 1.1: Author Publication Guidelines	83
Appendix 1.2: Search Terms	85
Appendix 1.3: Quality Appraisal Tool with Scoring Criteria	86
Appendix 1.4: Additional Demographic Information from Papers	90
Appendix 1.5: Additional Effect Sizes for Articles	92
Major Research Project	
Appendix 2.1: Author Publication Guidelines	93
Appendix 2.2: Major Research Proposal (MRP) V.5	95
Appendix 2.3: Health and Safety Form (MRP V.5)	110
Appendix 2.4: Research Costs (MRP V.5)	113
Appendix 2.5: Plain English Summary (MRP V.5)	114
Appendix 2.6: Research Ethical Approval Letters	117
Appendix 2.7: Information Pack	123
Appendix 2.8: Consent and Assent Forms	127
Appendix 2.9: Interview Schedule	131
Appendix 2.10: Excerpt from Transcript Analysis	134

CHAPTER ONE: SYSTEMATIC LITERATURE REVIEW

Psychosocial Adaptation in Parents of Children with Cardiac

Arrhythmias: A Systematic Review

Louisa Casselden, MA (Hons), MSc

Institute of Health & Wellbeing

College of Medical, Veterinary and Wellbeing

University of Glasgow

Word count: 8043

July 2015

Requests should be addressed to: Louisa Casselden, Gartnavel Royal Hospital, Administration

Building, 1st floor, 1055 Great Western Road, Glasgow G12 0XH, Scotland, UK

l.c.young.1@research.gla.ac.uk

Written in accordance with the requirements for submission to British Journal of Health

Psychology (Appendix 1.1)

Abstract

Purpose: This review aims to summarise the recent evidence base for psychosocial adaptation in parents of children with cardiac health conditions. The secondary aim is to compare differences in the psychosocial adaptation of mothers and fathers in this population.

Methods: The databases Embase, Medline, PsychInfo, CINAHL and Web of Knowledge were searched using a systematic search strategy. Inclusion criteria were: quantitative studies, English language only, parents of children with cardiac health difficulties, peer reviewed articles, published between 2004-2014, studies reporting parental mental health using standardised tools. Additional articles were identified by hand searching. A quality appraisal tool was constructed to evaluate the quality of papers.

Results: Parents of children with congenital heart disease (PCCHD) are at risk of psychosocial morbidities including increased anxiety, depression and stress. These difficulties are chronic for a significant proportion. Parental stress appears to be related to specific child health related events. Findings suggest that stress is heightened during specific events then reduces over time and is subsequently comparable to normative groups. Mothers reported significantly higher levels of psychosocial distress than fathers and there were gender differences in coping strategies. Limitations and clinical implications of the findings are discussed.

Conclusion: Parents of children with CHD are at heightened risk of psychosocial morbidities which may persist over time. Services need to identify families at risk in order to provide appropriate support and information. More research with fathers is required.

Introduction

At present, one in 180 children are born with a congenital heart disease (CHD) in the UK (Townsend et al., 2013). Treatment for cardiac conditions can range from regular medical monitoring for mild conditions, to repeated invasive surgery and frequent hospital admissions for more severe conditions (NHS Commissioning Board, 2013). As increasing numbers of children and young people with these life-threatening conditions are surviving and living for longer (Townsend et al, 2013), paediatric cardiac conditions are more frequently being managed as chronic illnesses. As a result, there is a developing need to explore the longer term impact of cardiac health problems on adjustment and quality of life in young people and their families (Bellinger & Newburger, 2010; Goldbeck & Melches, 2005).

Chronic illness places an ongoing strain on both children and their parents, and a process of re-adjustment is required at different stages of the child's development (Wallander & Varni, 1998). In addition, normal daily activities may be repeatedly disrupted due to the impact of treatment burden and physical constraints placed on the child by the illness. One meta-analysis of 87 studies by Lavigne & Faier-Routeman (1992) suggests that children with chronic illnesses are at higher risk of poor adjustment to their health condition and poorer mental health than general controls, although the majority of this population do have positive outcomes (Eiser, 1990).

Models of stress and coping have been developed to identify factors which impact on a young person's adjustment to a chronic illness. These have identified social-ecological factors such as maternal adjustment as a key factor in child adjustment to

their health condition. Thompson and Gustafson's model (1996) postulated that maternal appraisals and coping style either protect the child from or exacerbate the inherent stress which a child experiences from the illness. Wallander and Varni (1995)'s generic illness model suggests that psychosocial stress is one risk factor for maladjustment in mothers (Wallander & Varni, 1995). In one study, mothers of 457 children with chronic health problems reported more negative affect and both parents reported more mental health treatments compared to parents of 1270 healthy children (Cadman, Rosenbaum, Boyle & Offord, 1991). There is, however, inconsistent evidence regarding reported maternal adjustment in mothers of children with different health conditions (Wallander & Varni, 1998), and a historical paucity of research with fathers in this population.

In terms of outcomes for parents of children with congenital heart disease (PCCHD), one review of the literature explored parental satisfaction with paediatric care of children with CHD and parent wellbeing across 80 studies (Lawoko, 2007). This review indicated that parents experience a higher degree of psychosocial morbidity than parents of children with other chronic health conditions (e.g. cystic fibrosis) and are more likely to suffer symptoms of hopelessness, depression and anxiety than parents of healthy children. The majority of the papers in this review were cross-sectional and therefore did not explore the adjustment of families over time. The current review does not focus on parental satisfaction (Lawoko, 2007), rather it seeks to provide an overview of the recent literature on psychosocial adjustment for PCCHD.

In recent years there have been significant advancements in the treatment of children with CHD and improvements in care available to families, which may affect the difficulties families present with. Qualitative research exploring the experience of families living with children with CHD (Bruce, Lilja & Sundin, 2014; Connor et al., 2010) revealed that families receiving higher levels of support and person-centred care were more likely to adapt positively to parenting a child with CHD, while complexity of child's disease and parental socio-economic status was linked with increased parental stress and family burden. There continues to be a scarcity of literature on the psychosocial adaptation of fathers to having a child with a chronic illness (Wallander & Varni, 1998), although evidence does suggest that fathers are impacted by having a child with CHD. For example, one recent study by Bright et al., (2013) suggested that having an infant with CHD had a mixed impact on father's attachment, with some feeling closer and others more reserved towards the child. It is vital to increase the evidence base specifically for this population in order to understand father's psychosocial adaptation, as well as the impact it has on the rest of the family system.

Rationale and objectives

The primary aim of this review is to summarise the recent evidence base for psychosocial adaptation in parents of children with cardiac health conditions. The secondary aim is to compare psychosocial adaptation of mothers and fathers, to extend the literature base for fathers in this population. This will aid our understanding of the family system adjustment which impacts on child adaptation to living with a cardiac health problem. Findings may inform development of support

for families of children with cardiac illness, to optimise the recovery of the child and their families.

Method

The procedure for analysis, search strategy and eligibility criteria were decided prior to commencing the literature search for this study. Methods are reported following PRISMA-P guidelines on reporting systematic literature reviews (Moher et al., 2015)

Eligibility Criteria

Prospective studies which assessed the psychosocial adaptation of PCCHD using standardised assessment tools were included in this review. No papers from Lawoko (2007) are included as those published in 2004 did not meet the other inclusion criteria for this review.

Limits were set to include quantitative studies, English language only, peer reviewed articles and papers published between 2004- 2014. This time frame was chosen to capture literature published over the last decade to reflect the most up to date picture for psychosocial adaptation in PCCHD. Studies involving parents of children and adolescents (<20 years) with cardiac health difficulties were included. Primary outcome measures were aspects of parental stress or mental health assessed using standardised tools.

Exclusion criteria included: qualitative studies, conference abstracts, reviews and dissertations. Studies reporting only Quality of Life (QoL) measures were excluded as these did not provide enough information regarding parental mental health.

Intervention trials were excluded in order focus on establishing an overview of parental mental health. An evaluation of interventions at this stage was considered out-with the scope of this literature review.

Information Sources and Search Strategy

The following electronic databases were searched: Embase, Medline, PsychInfo, CINAHL and Web of Knowledge on 3/01/2015. Search terms varied slightly on different databases as a result of MeSH headings and Boolean operators. See Appendix 1.2 for full search terms. The following search terms were used:

[mental disorders/ or coping behaviour/ or mood disorders/or ((adaptation or stress or support or well being or resilience or adjustment) adj3 psycholog*)]

AND

[(parent* or parenting or parent-child relations or family) adj3 relation*]

AND

[(cardiac or cardio* or heart) adj4 (arrhythmi* or disease or death or surgery or surgical)) or (artificial pacemaker or implantable defibrillator or cardioverter)].

Study Selection

The electronic search generated 670 articles. Once duplicates were removed, titles and abstracts were screened in accordance with the inclusion and exclusion criteria identified below, and unsuitable articles were excluded. The references of the remaining 17 articles were reviewed by hand-searching and eight further articles were identified. Of these 25 potential articles, ten were excluded (Figure 1). The final 15 papers were reviewed systematically.

Data Collection

The final fifteen papers were reviewed using a quality assessment tool which the researcher constructed with reference to the Scottish Intercollegiate Guidance Network (SIGN) Methodology Checklist 3: Cohort Studies (Sleith, 2012) and Crowe Critical Appraisal Tool (Crowe, 2013). The quality assessment tool (See Appendix 1.3) was piloted on four papers and then reviewed. The quality appraisal tool rates papers on eight areas: Title and Abstract, Introduction, Design, Sampling, Data Collection, Ethical Issues, Analysis and Discussion. It includes twenty five items, rated as: Well Covered (2 points), Adequately Covered (1 point), Not Covered/Poorly Covered (0 points), with a final score out of 50. Assessment of risk of bias, management of attrition and consideration of confounding variables in the design, analysis and discussion of findings for each individual study were rated as part of the quality assessment tool.

An eligibility assessment was carried out with a sample of 6 papers, which were rated independently by two doctorate trainees and checked for inter-rater reliability. Where there were discrepancies within rating, these items were discussed until an agreement was reached. This happened with two items during the process: a) identifying confounding variables and b) rating ethical concerns. A final agreement on how these should be scored was reached, and the researcher applied this marking scheme to all papers in the review.

Results

Fifteen included studies are summarised in Table 1.

Demographic information

The fifteen papers reviewed had a total of 3072 participants. Twelve studies specified parent gender, 1389 (68%) of the responses from mothers and 670 (32%) from fathers. Fourteen papers specified number of children (n= 1733). Parental ages ranged from 20-44 years for mothers and 28-50 years for fathers. Child ages ranged from 1 day to 17 years. Details of cardiac condition and level of parental education are detailed in Appendix 1.4.

Three studies explored psychosocial functioning in children, however, also used measures to capture parental stress and mental health constructs therefore were included in this review (Brosig et al., 2007; Landolt et al., 2014; Majnemar et al., 2006).

Figure 1: Flow Diagram of Selection Process

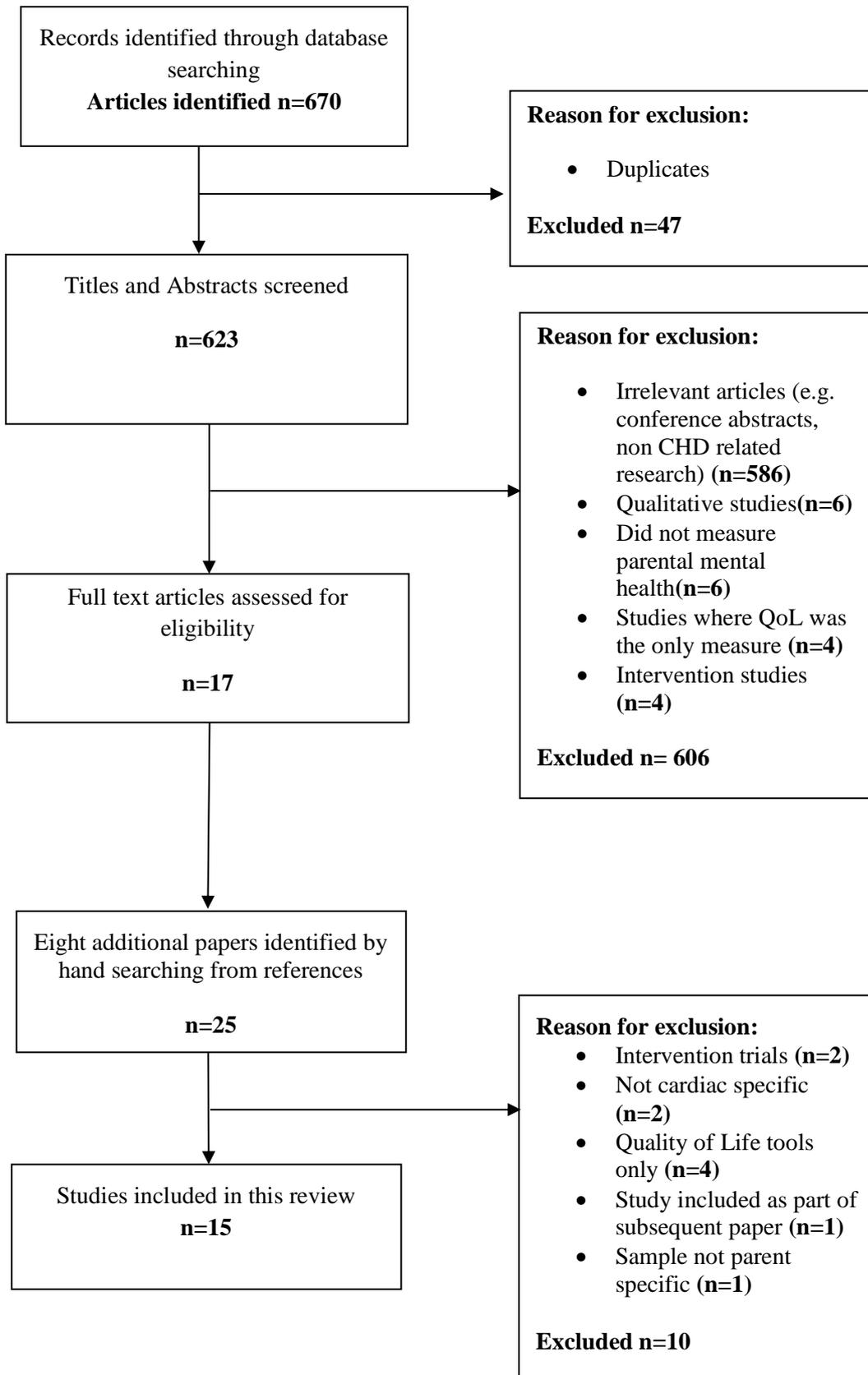


Table 1: Overview of Studies

Authors, year, country	Primary Aim	Comparisons	Participants	Design	Measures used	Data Analysis	Main findings	Quality rating (%)
Berant, Mikulincer & Shaver, (2008) Israel	To explore the contribution of mother's attachment style to child mental health (MH)	No control group Within subjects.	63 mothers of children diagnosed with CHD in their first year of life. The first 2 time points had 85 mothers included.	Longitudinal study. Within subjects design. Measures were assessed at time of diagnosis (T1), 1 year later (T2) and 7 years later (T3)	Mikulincer, Forian & Tolmacz's attachment scale Mental Health Inventory Evaluating & Nurturing Relationship Issues Communication and Happiness Scales (Hebrew)	Within-subjects repeated measures ANOVA Regression analysis Pearsons Correlation	Maternal avoidant attachment was the best predictor of deterioration in mother's MH and marital satisfaction over 7 years, particularly for those with children with severe CHD. Mother's attachment insecurities were associated with children's emotional problems and poor self-image 7 years later.	80
Brosig, Mussatto, Kuhn & Tweddell. (2007) North America	To assess the psychosocial outcomes for pre-school children & their families after surgery for complex CHD.	Parents of children with hypoplastic left heart syndrome (HLHS) Parents of children with transposition of the great arteries (TGA) Normative sample of healthy children.	13 children with HLHS, 13 with TGA. Test norms for healthy sample.	Prospective, cross sectional study. Measures assessed once	Pediatric Quality of Life Inventory Impact on the family Scale Parenting Stress Index Parent Behaviour Checklist Child Behaviour Checklist	Mann-Whitney Spearman Rank Correlation	No statistically significant differences in the Peds QL. Effect sizes for physical/emotional domains may be clinically meaningful. Parents of cardiac groups reported higher QoL than parents of children with other chronic illnesses. Parents of children with HLHS report higher levels of stress and more negative impact of all subscales of impact on the family. Benefits of having a child with CHD: closer family, feeling better about themselves by managing illness. Parenting stress correlated positively with parental perception of total impact of the illness on the family.	86

Authors, year, country	Primary Aim	Comparisons	Participants	Design	Measures used	Data Analysis	Main findings	Quality rating (%)
Doherty et al. (2009) UK	To examine psychological functioning and coping styles in mothers and fathers in the early months of life of an infant with severe CHD.	Comparing mothers and fathers of infants with severe CHD.	Parents of 70 infants.	Prospective study, cross sectional study Measures assessed once.	Brief Symptom Inventory Carvier, Scheier & Weintraub multidimensional coping inventory Maternal worry scale Family environment scale Significant Others scale Cardiac symptom checklist	MANCOVA Mann-Whitney between group comparison Regression analysis	Clinically elevated psychological distress in 1/3 of mothers and 1/5 of fathers in the months after birth of child with severe CHD. Mothers and fathers used different strategies to cope. Mothers had significantly higher levels of psychopathology compared with fathers. Coping-behaviour disengagement/ understanding diagnosis/ family-cohesion variables explained 44% of variance for mothers mental health. Worry and coping-use of alcohol were significant for fathers.	82
Franck, Mcquillan, Wray, Grocott & Goldman. (2010) UK	To investigate pre- & post-operative parental stress & to examine factors during post-operative period for children undergoing cardiac surgery	Two cohorts of parent of children with CHD	Parents of 211 children who have undergone cardiac surgery	Two phase prospective, within group repeated measures descriptive design. Measures assessed on days 3, 5, 8 & 15.	Pediatric Index of Mortality 2 Risk Adjustment in Congenital Heart Surgery-1 Parent Stressor Scale: Infant Hospitalisation Index of Multiple Deprivation		Parental stress remained moderate to high throughout child's hospitalisation, regardless of severity of illness. There were few differences between mother and fathers' stress, or perceptions of the illness. Parents in more deprived communities and mothers born outside of the UK had higher stress levels.	72

Authors, year, country	Primary Aim	Comparisons	Participants	Design	Measures used	Data Analysis	Main findings	Quality rating (%)
Helfricht, Latal, Fischer, Tomaske & Landolt. (2008) Switzerland	To evaluate post-traumatic stress disorder in parents of children who have undergone cardiopulmonary bypass surgery.	Mothers/ fathers of children with CHD	135 mothers & 98 fathers At six month follow up: 122 mothers & 92 fathers	Prospective cohort study. Measures assessed within 2 weeks of diagnosis (T1), at 2 weeks (T2) & at 18 months (T3)	Posttraumatic Diagnostic Scale	Paired Student's T-tests Bivariate Pearson's correlations	After discharge 16.4% of mothers and 13.3% of fathers met criteria for acute PTSD, while another 15.5% of mothers and 13.3 % of fathers had significant symptoms of post-traumatic stress. At six month follow up PTSD rates were 14.9% and 9.5% respectively.	82
Hendriks et al. (2005) Netherlands	To assess levels of specific distress and general distress 18 months in parents after their child having received the diagnosis of long QT syndrome	Parents of non-carrier children	36 parents (17 couples and 2 single parents) 41 children, 17 of which were carriers.	Prospective, longitudinal study. Measures assessed after consultation, at 2 weeks and 18 months.	Impact of events scale Spielberger State Anxiety Inventory Beck depression inventory Social Support Questionnaire Utrecht Coping List Semi structured interview	Independent t-tests Mann-Whitney tests Wilcoxon matched pairs tests Restricted Maximum likelihood estimation Chi square Univariate analysis	Parents of carrier children reported more distress than parents of non-carrier children, even at 18 months. Those with high disease related anxiety scores and those with low education, those more familiar with the syndrome and those who had experienced a sudden death in the family, and those who were unsatisfied with the level of information given had higher levels of distress.	70

Authors, year, country	Primary Aim	Comparisons	Participants	Design	Measures used	Data Analysis	Main findings	Quality rating (%)
*Landolt, Ystrom, Stene-Larsen, Holmstrom & Vollrath. (2014) Norway	To assess child internalising behaviour problems and maternal mental distress in a sample with CHD in first 3 years of life and the factors impacting these.	Controls from normative sample from the Norwegian Mother and Child Cohort study (MoBa).	408 children with CHD. MoBa normative sample of n=93009	Prospective, longitudinal study. Measures assessed at 6mths, 18mths and 36mths.	Infant Characteristics questionnaire-Fussy/Difficult scale Child Behaviour Checklist Hopkins Symptom Checklist	Structural equation modelling Poisson regression analysis	Elevated levels of maternal distress for mothers of children with CHD at all three assessments, with mothers of children with severe CHD at highest risk of problems. Increased levels of internalising behaviour problems in children with CHD. CHD contributed on average to 31% and 39% of variance in children's and mothers' problems respectively.	86
Lawoko & Soares (2006) Sweden	To assess long term psycho-social morbidity and its determinants among parents of children with congenital heart disease	Comparison of mothers & fathers	632 parents of children with CHD	Prospective, Longitudinal cohort study. Measures assessed at 1 time point & then 1 year later	Symptom checklist-Revised Hopelessness Scale Schedule for Social Interaction Pyramid patient questionnaire Client satisfaction questionnaire Financial questions	Independent sample t-tests Paired sample t-tests Chi-square tests Blockwise logistic regression analysis Fishers exact test	Prevalence of depression, anxiety, somatization & hopelessness symptoms among PCCHD was high at T1 & T2. Clinical severity did not significantly predict parents' psychosocial picture. Parent's perception of child's hospital care and perceived burden of care over time were significant determinants of morbidity. Mothers were more likely to report psychosocial symptoms and had higher prevalence of these. Improved social network was associated with better psychosocial status.	84

Authors, year, country	Primary Aim	Comparisons	Participants	Design	Measures used	Data Analysis	Main findings	Quality rating (%)
Majnemar et al. (2006) Canada	To determine health related quality of life in young children with CHD, and levels of parental stress 5 years after surgery.	Normative sample	49 parents of children with CHD	Prospective, longitudinal design. Measures assessed at time of open heart surgery (detailed in previous study) & then 5 years later	Child Health Questionnaire Parenting Stress Index	Correlation Chi Square analysis T tests Simple and multiple linear regression	Parental stress at follow up increased by a factor of 4.5 if their child was cyanotic prior to the first open heart surgery. Child behaviour at follow up was also significantly correlated with parental stress. Parental distress, helplessness and other stresses were associated with overall family wellbeing. Severity of the defect or level of disability, were not important predictors.	68
Menahem, Poulakis & Prior. (2008) Australia	To investigate psychological & emotional experiences of parents of children undergoing cardiac surgery.	Normed samples for standardised tests.	29 mothers and 28 fathers of children with CHD (only 10 fathers responded)	Prospective, longitudinal study. Measures taken prior to surgery and 12-50 months follow up	State-trait anxiety inventory General Health Questionnaire Levenson's locus of control questionnaire Family assessment device Index of social support	Pearson product-moment correlations	Reported increase of maternal anxiety at the time of surgery was reported as diminished by 12 months or later. Ratings of MH difficulties were significantly reduced from time of surgery to 12 months or later.	44

Authors, year, country	Primary Aim	Comparisons	Participants	Design	Measures used	Data Analysis	Main findings	Quality rating (%)
*Solberg et al. (2012) Norway	To investigate the association between infants' congenital heart defects (CHD) and compromised maternal mental health	Different severities of child CHD, healthy controls from normative sample from the Norwegian Mother and Child Cohort study.	141 mothers of children with CHD 36,437 mothers from the Norwegian mother and Child Cohort study.	Prospective, longitudinal study. Measures taken at 6mths, 18mths and 36mths.	Hopkins Symptoms Checklist-25	Mixed between-within subjects analysis of covariance Cross sectional analysis for 36mths measures. Chi squared & 2 sample t-tests	Mothers with children with severe CHD have significantly higher depression and anxiety symptoms than other CHD groups and controls, even at 36 months post-partum.	94
Spijkerboer et al. (2007) Netherlands	To assess psychological distress & styles of coping in mothers & fathers of children undergoing invasive treatment for congenital cardiac disease	Parents of children with 4 different cardiac diagnoses. Normative sample from standardised tool manuals	52 sets of parents, 48 mothers and 9 fathers on their own (GHQ) 53 sets of parents, 50 mothers and 6 fathers alone (UCS)	Longitudinal design, cross sectional. Measures assessed 7 years & 6 months after invasive cardiac surgery.	Utrecht Coping List The General Health Questionnaire	One sample t-test Paired t-test univariate analysis of covariance	Parents of children with CHD showed lower levels of distress, e.g somatic symptoms, anxiety, sleeplessness & depression. Mothers of children with CHD reported significantly more somatic symptoms than fathers. Mothers tended to seek more social support than fathers. More favourable outcomes for coping were found: less expressed negative emotions and a weaker tendency to use reassuring thoughts.	80

Authors, year, country	Primary Aim	Comparisons	Participants	Design	Measures used	Data Analysis	Main findings	Quality rating (%)
Torowicz, Irvine, Hanlon, Sumpter & Medoff-Cooper. (2010) North America	To identify and compare differences in temperament and maternal stress between infants with complex congenital heart disease and healthy controls at 3 months of age.	Mothers of healthy controls, single ventricle and bi-ventricle groups.	33 mothers of children with single ventricle disease, 36 mothers of children with bi-ventricle disease 60 mothers of healthy children.	Cross-sectional design. Measures taken once. Part of a larger longitudinal study.	Early Infancy Temperament Questionnaire Parenting Stress Index, long form	Chi square One way analysis of variance Tukey's tests Pearsons correlation coefficients	Mothers of infants with SV physiology are more stressed than mothers of children with BV physiology on child domain subscales, summary scales and Life Stress subscale. None of the CHD mothers had higher levels of depression. Negative mood and high distractibility mutually explain >50% variance for total stress subscale	68
Vrijmoet-Wiersma, Ottenkamp, van Roozendaal Grootenhuis & Koopman (2009) Netherlands	To describe levels of parental stress & perceived vulnerability in parents of children who have undergone major cardiac surgery.	Normative samples for each instrument	114 mothers and 82 fathers of 131 children	Cross-sectional design. Measures assessed once.	Pediatric Inventory for Parents, short form. General Health Questionnaire Parental Stress Index Short Form State-Trait Anxiety Index Child Vulnerability Scale	Independent t-tests ANOVA Forced entry regression analysis	Levels of distress in parents of children with congenitally malformed hearts (PCCMH) were comparable to normative groups. State anxiety was higher in mothers of children with CMH. Significantly higher parenting stress and perceived vulnerability was found in PCCMH than in normative samples.	80

Authors, year , country	Primary Aim	Comparisons	Participants	Design	Measures used	Data Analysis	Main findings	Quality rating (%)
Wray & Sensky, (2004) UK	To assess levels of distress, the marital relationship & styles of coping of parents of children with congenital heart disease in comparison to two other groups.	Parents of children undergoing another hospital treatment (bone marrow transplant), & parents of healthy children.	Cardiac group: 54 mothers & 48 fathers (n=102) Bone marrow group (BMT): 67 mothers, 47 fathers (n=114) Healthy group: 66 mothers, 56 fathers (n=122)	Prospective, longitudinal study. Measures assessed pre & post operation, then 12 month follow up.	General Health Questionnaire Dyadic Adjustment scale Utrecht coping list Locus of control scale devised for the study.	Chi Square McNemar Chi Squared One way ANOVA Scheffe's multiple comparison tests Independent & paired t-tests Correlation coefficients	Parents of groups undergoing surgery reported significantly higher levels of distress than healthy controls, which reduced after surgery. No significant differences in distress between groups undergoing surgery, or between different severity of cardiac problems. Higher degree of marital satisfaction reported for mothers of cardiac children than BMT group after surgery. Strategies for coping changed over time for mothers of BMT children and fathers of cardiac children.	84

**Landolt et al., (2010) and Solberg et al., (2012) used the Norwegian Mother and Child Cohort Study database (MoBa) . The MoBa study participation rate is 38.5%, and the comparison groups were n=36,437 and n=93,009 for the Solberg (2012) and Landolt (2014) papers respectively*

Research Design

Ten of the studies in this review utilised a longitudinal, repeated measures design to explore the psychosocial adaptation of PCCHD over time. Measures were repeated at 12, 18 or 36 months after initial assessment, one followed up PCCHD up to five years (range 12 months-50 months) after surgery (Menahem et al., 2007) and two others at seven years (Berant et al., 2008; Spijkerboer et al., 2007).

Five papers used a cross-sectional design (Brosig et al., 2007; Frank et al., 2010; Torowicz et al., 2010; Vrijmoet-Wiersma, 2009). While this design is useful to provide a snapshot of parental adjustment at specific time points, it does not reflect changes in adjustment over time or causality. One study reported a power calculation, and this was high at > 0.9 (Doherty et al., 2009), indicating that findings are less likely to be affected by Type 1 or Type 2 error. Many of the studies did not report power calculations or effect sizes, or noted a lack of power as a weakness in the study (Brosig et al., 2007; Majnemar et al., 2006). See Appendix 1.5 for additional effect size calculations.

Quality Rating

Using the quality tool designed for this review, ten papers scored $>80\%$ (see Table 3). This suggests they were of high quality and sufficiently detailed in their reporting to allow confidence in their interpretation of findings. Four papers scored between 60-80%, with scores affected by: confounding variables not accounted for, lack of information regarding attrition and missing data, risk of bias from inequalities in data collection procedures, reporting p values only without confidence intervals, effect sizes or power. One paper (Menahem et al., 2008) was rated as

44%, indicating that caution should be used when considering the findings as this study did not meet several of the quality assessment criteria.

Findings

To synthesise the recent evidence base for psychosocial adaptation in parents of children with cardiac health conditions, the psychosocial outcomes for parents are first reviewed and the factors associated increased risk of psychosocial morbidity are discussed, followed by the evidence of outcomes for fathers.

Psychosocial constructs

Psycho-social constructs such as parenting stress, depression and anxiety were commonly explored. Other constructs such as marital status coping, family cohesion and functioning, locus of control, social support and post-traumatic stress were also investigated. Evidence for levels of distress in PCCHD varied between papers depending on the construct being measured.

Overall, studies suggested that parents of children with severe CHD have significantly higher levels of anxiety and depression than parents of children with mild or moderate CHD, (Vrijmoet-Wiersma et al., 2009; Menahem, et al., 2008; Hendricks et al, 2005; Doherty et al., 2009). One study also found that a significant minority of PCCHD showed symptoms of post-traumatic stress (Helfricht et al, 2008). Difficulties were found to persist up to 18 months later (Solberg et al., 2012; Lawoko & Soares, 2006; Helfricht et al, 2008). Landolt et al. (2014) found that parental distress in PCCHD was significantly higher than the normal population and tended to increase over time, however there was also an increase in child

internalising problems and fussiness over this time, making it difficult to establish causality. Effect sizes for this study were low ($d= 0.17-0.28$), suggesting that the clinical implications of these findings may be limited.

Some papers identified differences in parental distress related to the type of CHD condition their child had (Vrijmoet-Wiersma et al., 2009; Torowicz et al., 2010) due to the increased treatment burden for children with more severe cardiac difficulties (Brosig et al., 2007). Others suggested that there was no difference in distress related to the type or severity of cardiac abnormality (Menahem et al., 2008; Doherty et al., 2009). Berant et al., (2008) found that mothers of children with severe CHD, who had an avoidant attachment style, had the largest deterioration in mental health over seven years compared with parents of children with mild to moderate CHD. The lack of control group in this study means it is unclear whether this finding is specific to PCCHD and does not address confounding variables which may have impacted on findings. Other studies found significantly higher parental stress in PCCHD than in normative samples or healthy controls (Vrijmoet-Wiersma et al., 2009; Torowicz et al., 2010; Majnemer et al., 2006).

Conflictingly, Spijkerboer et al. (2007) found that levels of distress were largely comparable to normative groups and reported lower levels of somatic symptoms and sleeplessness. In this study, one in five parents did not respond, and findings are at risk of bias if more distressed families were less likely to take part. Stress seemed significantly higher for parents during times of acute treatment such as surgery however parents reported that stress reduces over time and is subsequently comparable to normative groups (Wray & Sensky, 2004, Franck et al., 2010;

Vrijmoet-Wiersma et al., 2009), or lower (Spijkerboer et al., 2007; Brosig et al., 2007).

Factors associated with psychosocial morbidity

A number of factors were identified which were associated with higher psychosocial morbidity, including: socioeconomic deprivation and mothers who were foreign nationals (Franck et al., 2010; Helfricht et al., 2008), characteristics of medical procedures (Vrijmoet-Wiersma et al., 2009); perceived lack of information and low parental education (Hendricks et al., 2005). Negative parental perceptions were found to be significant determinants of psychosocial morbidity over time, such as impressions of impact of the illness (Brosig et al., 2007), hospital care, perceived burden of care and financial burden (Lawoko & Soares, 2006; Doherty et al., 2009; Majnemer et al., 2006). A number of medical factors were not found to correlate with parental psychosocial adaptation, such as severity of conditions (Lawoko & Soares, 2006) or length of stay in intensive care (Franck et al., 2010). Improved social network was associated with improved psychosocial status and parental distress, helplessness and other stresses were associated with overall family wellbeing (Lawoko & Soares, 2006; Majnemer et al., 2006). Doherty et al. (2009) found that mental health difficulties were significantly associated with knowledge, appraisals, coping and family functioning, but found no significant associations between socio-economic status or social support.

Comparisons of mothers and fathers

Findings indicate that there are significant differences between mothers and fathers in the presentation of distress and coping styles. Six studies which compared gender

are summarised in Table 2. In all studies male and female respondents were recruited from the same families.

Table 2: Comparisons of Mothers and Fathers

Findings	Article
Mothers of children with severe CHD showed significantly higher levels of higher generic stress, parenting stress, anxiety and perceived vulnerability scores than fathers	Vrijmoet-Wiersma et al., (2009)
Mothers showed a higher incidence of clinically elevated psychological distress than fathers	Doherty et al., (2009); Lawoko & Soares, (2006); Helfricht et al.,(2008)
Mothers more likely than fathers to report psychosocial symptoms over time	Lawoko & Soares, (2006)
Mothers reported more somatic symptoms than fathers	Spijkerboer et al., (2007); Helfricht et al., (2008)
Parents of children with severe CHD were found to use different coping strategies. Mothers were more likely to vent, draw on social support (instrumental and emotional) and religious/spiritual support while fathers used alcohol significantly more as a coping strategy.	Doherty et al., (2009); Spijkerboer et al., (2007)
Mothers reported significantly higher symptom severity of PTSD at discharge and at 6 month follow up than fathers. No significant differences in the frequency of PTSD in either within a month of surgery or at 6 month follow up. Mothers were more likely to be younger, single, unemployed and on sick leave than fathers and had more difficulty raising money. The length of time spent caregiving to CCHD decreased significantly for fathers over time, but did not for mothers.	Helfricht et al, (2008)
Few differences between mother and fathers stress, or their perceptions of the illness	Franck et al (2010)

Finally, some positive outcomes of having a child with CHD were identified. Higher marital satisfaction (Wray & Sensky, 2004), higher quality of life for the family and less negative impact than parents of children with other chronic illnesses (Brosig et al., 2007) were identified in PCCHD than comparison data.

Discussion

Overall the evidence suggests that parents of children with congenital heart disease are at higher risk of developing a range of psychosocial morbidities than the general population. This is consistent with previous findings from a literature review by Lawoko (2007) and with evidence from parents of children with other chronic health conditions (Cadman et al., 1991). The longitudinal studies in this review suggest that anxiety and depression are chronic for a significant proportion of PCCHD. In contrast, parental stress appears to be related to specific life events. As may be expected, stress seems to be a transient state which is experienced as highest around surgical interventions. In the longer term parents either adapt successfully to having a child with CHD, or continue to report symptoms of clinically significant psychosocial morbidities. The research suggested that parental coping strategies change over time, which may reflect parental adjustment to living with a child with CHD.

A number of papers in this review identified significant differences between mothers and fathers of CCHD, as females reported higher levels of psychosocial distress than males (Doherty et al., 2009; Helfricht et al., 2008; Lawoko & Soares, 2006; Vrijmoet-Wiersma et al., 2009). This finding may reflect higher prevalence rates of anxiety and mood disorders in women (Seedat et al., 2009). In addition, females

may also be more likely to identify symptoms of distress and seek support than males. There were significant differences between fathers and mothers coping strategies (Doherty et al, 2009), with females drawing on more social and spiritual support while fathers were more likely to use alcohol. This may reflect that men are consistently found to experience higher rates of substance abuse and externalising disorders than women (Seedat et al., 2009). It may also suggest a wider cultural ethos where it is more acceptable for women to express distress and access support than men, not that fathers do not experience distress in relation to caring for a child with CHD.

Parental perceptions of their child's illness are a significant aetiological factor in psychosocial distress. This concurs with the literature base for models of stress and coping which highlight the importance of maternal appraisals of their child's chronic illness (Wallander & Varni, 1995; Thompson & Gustafson, 1996). The evidence for severity of the illness as a factor in psychological distress is mixed as several studies found that severity of CHD is not linked with the level of clinical distress that parents report (Lawoko & Soares, 2006), while others showed that parents of children with severe CHD were more likely to experience significant anxiety and depression than parents of children with mild to moderate difficulties (Vrijmoet-Wiersma et al., 2009). Other factors that affected psychosocial adaptation were social support, other family stressors such as socio-economic deprivation and also whether the parents were foreign nationals. Mothers of CCHD who were foreign nationals reported higher levels of distress. One explanation for this may be that these families have less established support networks than families where the treatment is taking place in the maternal country of origin. Alternatively, distress

may be higher where mothers have less familiarity with the medical system, or where they may be less able to access information in their native language. Hendricks et al., (2005) suggested that distress was higher where there was a perceived lack of information, which may be exacerbated where information is provided in a different language.

Methodological limitations and future research

The generalisability of the findings this review are limited by a number of factors. Only one paper provided a power calculation (Doherty et al., 2009) to justify the sample size, making difficult to rule out the risk of Type II error in other studies due to lack of power. Many of the findings, however, were replicated in different studies, suggesting this risk was not high. All papers utilised self-report measures, which do have inherent difficulties such as risk of biased reporting and acquiescence. Future studies would benefit from corroboration of self-report measures with other sources of information such as a structured clinical interview or observation to strengthen findings and reduce risk of bias. The papers in this review were all conducted in high- to middle- income countries and it would be useful to expand the scope of future research to developing countries where there may be different psychological and physical healthcare implications of CHD due to differences in healthcare provision, in order to increase the generalisability and cultural validity of findings.

As the population of children with CHD is small, it is often difficult to recruit sufficient numbers, particularly when categorised by type of CHD. As a result, participant experiences may vary in terms of hospital experience or treatment

burden, which affect perceived levels of distress. It may be harder to identify meaningful differences where there are large discrepancies between participants. For example, Menahem et al., (2007) followed up PCCHD between 12 months-50 months after surgery. This extended time-frame is problematic as multiple variables could impact parental adjustment at different times during their child's development, impacting on the validity of the findings and making it difficult to draw conclusions about this sample.

The evidence identified significant differences between mothers and father of CCHD and further research is necessary to strengthen the evidence base and increase our understanding of the differences and similarities in their psychosocial adaptation. Further research into the differences in attributions and coping styles in fathers and mothers would allow services to develop effective means of engaging fathers who may be struggling with psychosocial adaptation. Support and information packages which optimise engagement and positive outcomes for parents may have a significant impact on the psychological wellbeing of whole family.

Clinical Implications

The findings from this review add further support to previous research findings (Lawoko & Soares, 2002) that parents of children with congenital heart diseases are at an increased risk of a number of mental health difficulties. Based on the literature on stress and coping models (Wallander & Varni, 1998), parental adaptation to their child's CHD is likely to have a significant impact on the child's adaptation to their illness, and may have important implications for the child's subsequent development (Berant et al., 2008). For a significant number of parents, psychosocial difficulties

such as anxiety, depression and PTSD persist over time and may develop into chronic mental health problems (Lawoko & Soares, 2006; Solberg et al., 2012). It is crucial, therefore, that services have systems in place to review families over time and that they are able to identify families who are struggling to adapt to their child's illness and deliver appropriate interventions in a timely manner.

Due to the apparent event-specific nature of heightened parental stress, it may be that short term interventions on stress management would be useful for families experiencing high levels of stress prior to hospital admission or to coincide with particularly stressful procedures such as surgery. Stress management interventions have been shown to be effective in other clinical health populations such cancer patients (Jacobsen et al., 2002).

The evidence suggests that parental perceptions of the severity of their child's illness are associated with increased psychological distress, rather than actual severity of the illness. Therefore it is important that healthcare professionals are sensitive to parental concerns and are able to provide support which is tailored to the needs of families, regardless of the severity of CHD. Professionals must be aware that families may experience high levels of distress even where their child's illness is not in the severe range.

Finally, it seems that there are significant differences between mothers and father's psychosocial adaptation having a child with CHD. Historically, fathers have been harder to engage in research in this population, although the number of studies including data from fathers is encouraging. It is important that healthcare services

strive to engage fathers and well as mothers in assessment, research and interventions. Fathers may be less likely to attend appointments with their children, or less likely to identify and present with symptoms of distress to services; however, a significant proportion of them may also suffer from mental health difficulties as a result of having a child with CHD. They may be more likely to use solitary coping strategies to cope with their distress. Healthcare organisations should be aware of this and consider ways in which encourage fathers to engage more effectively with support services.

Conclusions

Overall, this review suggests parents of children with congenital heart diseases are at heightened risk of psychosocial morbidities which may persist over time. It is important, therefore, that services are able to identify the families who are at risk and they can provide appropriate interventions or signpost families to appropriate support and information. The evidence suggests that mothers are more likely to report symptoms of psychological distress than fathers; however more research is required to understand the reasons for this.

References

- Bellinger, D. C., & Newburger, J. W. (2010). Neuropsychological, psychosocial, and quality-of-life outcomes in children and adolescents with congenital heart disease. *Progress in Pediatric Cardiology*, 29(2), 87-92.
doi:10.1016/j.ppedcard.2010.06.007
- Berant, E., Mikulincer, M., & Shaver, P. R. (2008). Mothers' Attachment Style, Their Mental Health, and Their Children's Emotional Vulnerabilities: A 7 - Year Study of Children With Congenital Heart Disease. *Journal of Personality*, 76(1), 31-66. doi: 10.1111/j.1467-6494.2007.00479.x
- Bright, M. A., Franich-Ray, C., Anderson, V., Northam, E., Cochrane, A., Menahem, S., & Jordan, B. (2013). Infant cardiac surgery and the father–infant relationship: Feelings of strength, strain, and caution. *Early human development*, 89(8), 593-599. doi: 10.1016/j.earlhumdev.2013.03.001
- Brosig, C. L., Mussatto, K. A., Kuhn, E. M., & Tweddell, J. S. (2007). Psychosocial outcomes for preschool children and families after surgery for complex congenital heart disease. *Pediatric Cardiology*, 28(4), 255-262.
doi:10.1007/s00246-006-0013-4
- Cadman, D., Rosenbaum, P., Boyle, M., & Offord, D. R. (1991). Children with chronic illness: family and parent demographic characteristics and psychosocial adjustment. *Pediatrics*, 87(6), 884-889.
- Connor, J. A., Kline, N. E., Mott, S., Harris, S. K., & Jenkins, K. J. (2010). The meaning of cost for families of children with congenital heart

disease. *Journal of pediatric health care*, 24(5), 318-325.

doi:10.1016/j.pedhc.2009.09.002

Crowe, M. (2013). *Crowe Critical Appraisal Tool v1.4*

Doherty, N., McCusker, C. G., Molloy, B., Mulholland, C., Rooney, N., Craig, B., ... & Casey, F. (2009). Predictors of psychological functioning in mothers and fathers of infants born with severe congenital heart disease. *Journal of Reproductive and Infant Psychology*, 27(4), 390-400.

doi:10.1080/02646830903190920

Eiser, C. (1990). Psychological effects of chronic disease. *Journal of Child Psychology and Psychiatry*, 31(1), 85-98.

doi: 10.1111/j.1469-7610.1990.tb02274.x

Franck, L. S., Mcquillan, A., Wray, J., Grocott, M. P., & Goldman, A. (2010). Parent stress levels during children's hospital recovery after congenital heart surgery. *Pediatric Cardiology*, 31(7), 961-968.

doi: 10.1007/s00246-010-9726-5

Goldbeck, L., & Melches, J. (2005). Quality of life in families of children with congenital heart disease. *Quality of Life Research*, 14(8), 1915-1924.

doi: 10.1007/s11136-005-4327-0

Goldbeck, L., & Melches, J. (2006). The impact of the severity of disease and social disadvantage on quality of life in families with congenital cardiac disease. *Cardiology in the Young*, 16(1), 67-75.

doi: 10.1017/s1047951105002118

- Helfricht, S., Latal, B., Fischer, J. E., Tomaske, M., & Landolt, M. A. (2008). Surgery-related posttraumatic stress disorder in parents of children undergoing cardiopulmonary bypass surgery: a prospective cohort study. *Pediatric Critical Care Medicine*, *9*(2), 217-223. doi: 10.1097/PCC.0b013e318166eec3
- Hendriks, K. S., Grosfeld, F. J. M., Van Tintelen, J. P., Van Langen, I. M., Wilde, A. A. M., Van Den Bout, J., & Ten Kroode, H. F. J. (2005). Can parents adjust to the idea that their child is at risk for a sudden death?: Psychological impact of risk for long QT syndrome. *American Journal of Medical Genetics Part A*, *138*(2), 107-112. doi: 10.1002/ajmg.a.30861
- Jacobsen, P. B., Meade, C. D., Stein, K. D., Chirikos, T. N., Small, B. J., & Ruckdeschel, J. C. (2002). Efficacy and costs of two forms of stress management training for cancer patients undergoing chemotherapy. *Journal of Clinical Oncology*, *20*(12), 2851-2862. doi: 10.1200/JCO.2002.08.301
- Landolt, M. A., Ystrom, E., Stene-Larsen, K., Holmstrøm, H., & Vollrath, M. E. (2014). Exploring causal pathways of child behavior and maternal mental health in families with a child with congenital heart disease: a longitudinal study. *Psychological Medicine*, *44*(16), 3421-3433. doi: 10.1017/S0033291713002894
- Lavigne, J. V., & Faier-Routman, J. (1992). Psychological adjustment to pediatric physical disorders: A meta-analytic review. *Journal of Pediatric Psychology*, *17*(2), 133-157. doi: 10.1093/jpepsy/17.2.133

- Lawoko, S., & Soares, J. J. (2002). Distress and hopelessness among parents of children with congenital heart disease, parents of children with other diseases, and parents of healthy children. *Journal of Psychosomatic Research*, 52(4), 193-208. doi: 10.1016/S0022-3999(02)00301-X
- Lawoko, S., & Soares, J. J. (2006). Psychosocial morbidity among parents of children with congenital heart disease: A prospective longitudinal study. *Heart & Lung: The Journal of Acute and Critical Care*, 35(5), 301-314. doi: 10.1016/j.hrtlng.2006.01.004
- Lawoko, S. (2007). Factors influencing satisfaction and well-being among parents of congenital heart disease children: development of a conceptual model based on the literature review. *Scandinavian Journal of Caring Sciences*, 21(1), 106-117. doi: 10.1111/j.1471-6712.2007.00444.x
- Majnemer, A., Limperopoulos, C., Shevell, M., Rohlicek, C., Rosenblatt, B., & Tchervenkov, C. (2006). Health and well-being of children with congenital cardiac malformations, and their families, following open-heart surgery. *Cardiology in the Young*, 16(2), 157-164. doi: 10.1017/S1047951106000096
- Menahem, S., Poulakis, Z., & Prior, M. (2008). Children subjected to cardiac surgery for congenital heart disease. Part 2—Parental emotional experiences. *Interactive cardiovascular and thoracic surgery*, 7(4), 605-608. doi:10.1510/icvts.2007.171066
- Moher, D., Liberati, A., Tetzlaff, J., & Altman, D. G. (2009). Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement.

Annals of internal medicine, 151(4), 264-269.

doi:10.7326/0003-4819-151-4-200908180-00135

Moher, D., Shamseer, L., Clarke, M., Ghersi, D., Liberati, A., Petticrew, M., ... & Stewart, L. A. (2015). Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015 statement. *Systematic Reviews*, 4(1), 1. doi:10.1186/2046-4053-4-1

NHS Commissioning Board (2013). *2013/14 NHS Standard Contract For Paediatric Cardiac: Cardiology And Surgery*. NHS England E05/S/a. Retrieved from <http://www.england.nhs.uk/wp-content/uploads/2013/06/e05-paedi-cardi-cardi-surg.pdf> (last accessed 3rd June 2015).

Seedat, S., Scott, K. M., Angermeyer, M. C., Berglund, P., Bromet, E. J., Brugha, T. ... & Kessler, R. C. (2009). Cross-national associations between gender and mental disorders in the World Health Organization World Mental Health Surveys. *Archives of General Psychiatry*, 66(7), 785-795. doi:10.1001/archpsyc.57.1.21.

Sleith, C., (2012). *Scottish Intercollegiate Guidance Network (SIGN) Methodology Checklist 3: Cohort Studies*. Healthcare Improvement Scotland: Edinburgh

Solberg, Ø., Dale, M. T. G., Holmstrøm, H., Eskedal, L. T., Landolt, M. A., & Vollrath, M. E. (2012). Trajectories of maternal mental health: A prospective study of mothers of infants with congenital heart defects from pregnancy to 36 months postpartum. *Journal of Pediatric Psychology*, 37(6), 687-696. doi: 10.1093/jpepsy/jss044

Spijkerboer, A. W., Helbing, W. A., Bogers, A. J., Van Domburg, R. T., Verhulst, F. C., & Utens, E. M. (2007). Long-term psychological distress, and styles of coping, in parents of children and adolescents who underwent invasive treatment for congenital cardiac disease. *Cardiology in the Young*, *17*(6), 638-645. doi:10.1017/S1047951107001333

Thompson, R. J., & Gustafson, K. E. (1996). *Adaptation to chronic illness*. Washington, DC: American Psychological Association.

Torowicz, D., Irving, S. Y., Hanlon, A. L., Sumpter, D. F., & Medoff-Cooper, B. (2010). Infant temperament and parental stress in 3-month-old infants after surgery for complex congenital heart disease. *Journal of Developmental and Behavioral Pediatrics*, *31*(3), 202-208. doi: 10.1097/DBP.0b013e3181d3dea

Townsend, N., Bhatnagar, P., Wickramasinghe, K., Williams, J., Vujcich, D., & Rayner, M., (2013). *Children and young people statistics 2013*. London: British Heart Foundation.

Vrijmoet-Wiersma, C. M., Ottenkamp, J., van Roozendaal, M., Grootenhuis, M. A., & Koopman, H. M. (2009). A multicentric study of disease-related stress, and perceived vulnerability, in parents of children with congenital cardiac disease. *Cardiology in the Young*, *19*(6), 608-614.
doi: 10.1017/S1047951109991831

Wallander, J. L., & Varni, J. W. (1995). Appraisal, coping, and adjustment in adolescents with a physical disability. In J.L Wallander & L.J. Siegel (Eds) *Adolescent health problems: Behavioral perspectives*. New York: Guilford Press

Wallander, J. L., & Varni, J. W. (1998). Effects of pediatric chronic physical disorders on child and family adjustment. *Journal of Child Psychology and Psychiatry*, *39*(1), 29-46. doi: 10.1111/1469-7610.00302

Wray, J., & Sensky, T. (2004). Psychological functioning in parents of children undergoing elective cardiac surgery. *Cardiology in the Young*, *14*(2), 131-139. doi:10.1017/S1047951104002045

CHAPTER TWO: MAJOR RESEARCH PROJECT

An Exploration of Body Image and Self-Esteem in Adolescents with Implantable Cardioverter Defibrillators (ICDs): A Qualitative Study.

Louisa Casselden, MA (Hons), MSc

Institute of Health & Wellbeing

College of Medical, Veterinary and Wellbeing

University of Glasgow

Word count: 8610

July 2015

Requests should be addressed to: Louisa Casselden, Gartnavel Royal Hospital, Administration

Building, 1st floor, 1055 Great Western Road, Glasgow G12 0XH, Scotland UK

l.c.young.1@research.gla.ac.uk

*Written in accordance with the requirements for submission to International Journal of
Qualitative Studies on Health and Well-Being (Appendix 2.1)*

Plain English Summary

Implantable Cardioverter Defibrillators (ICDs) have been shown to have good outcomes for people who are at risk of life-threatening irregular heart rhythms, providing longer life expectancy and reducing the occurrence of heart failure. ICDs create small electric pulses which induce a regular heart rhythm. Research suggests that adult patients often experience on-going psychological, social and physical changes after getting an ICD, but evidence for children and adolescents with ICDs is limited. Body image issues may be particularly important for adolescents with visible health conditions, as they experience typical physical and emotional development in addition to the burden of their health condition. The aim of this study was to explore the development of body image and self-esteem in this group, and other challenges which they experience. Ethical approval for this project was given by the West of Scotland Research Ethics committee. Interviews lasting between thirty minutes and one hour were carried out with four males with ICDs, aged 12-17 years old at the Royal Hospital for Sick Children, Glasgow. Interviews were recorded and typed up word for word, with data removed that could identify participants.

The interview transcripts were read over multiple times and analysed for common themes. Six main themes were found: Physical effect, Emotional impact, Sense of self, Coping Strategies, Development over time and Evaluation of ICD. The findings suggested that ICDs influenced participants' sense of self, and for some had a negative effect on body image. Although participants identified some negative consequences of having an ICD, overall they viewed the ICD positively and showed adjustment to the ICD over time.

Abstract

Introduction. Adolescents with Implantable Cardioverter Defibrillators (ICDs) have to negotiate the tasks of growing up while coping with their ICD device. Current research on the psychosocial effect of ICDs in this population is limited. Issues of body image may be particularly salient for adolescents with visible health conditions, as they experience typical physical and emotional development in addition to the burden of their health condition. This study aims to explore the effect of having an ICD device on adolescent's body image and self-esteem, and other challenges they encounter.

Method. This study utilised a qualitative research design comprised of in-depth individual interviews lasting between thirty minutes to one hour. Participants were recruited from the Royal Hospital for Sick Children, Glasgow. The data were analysed using Interpretative Phenomenological Analysis (IPA).

Results. The sample comprised four males aged between 12-17 years old. Six superordinate themes emerged from the analysis of the transcripts: Physical effect, Emotional impact, Sense of self, Coping Strategies, Development over time and Evaluation of ICD.

Conclusions. The findings suggested that ICDs influenced participant's sense of self, and for some had a negative effect on body image. Although some negative consequences of having an ICD were identified, participants showed a determination to overcome challenges and a positive progression in adjustment over time.

Keywords: *Artificial heart pacemaker, cardiac arrhythmia, paediatric, children, Interpretative Phenomenological Analysis.*

Introduction

Children born with congenital heart disease (CHD) are at risk of a range of significant health problems, including arrhythmias of the heart, which can be fatal. Devices such as Implantable Cardioverter Defibrillators (ICDs) have been shown to be effective in treating potentially life threatening arrhythmias with both adults (NICE, 2006) and a paediatric population (Stefanelli et al., 2002). These ICDs are implanted under the collar-bone and create small electric pulses which induce a regular heart rhythm. The medical benefits of these devices have been documented, providing a moderately longer life expectancy for adults who receive an ICD compared to those who do not, reducing the occurrence of heart failure and sudden cardiac arrest (Bardy et al., 2005). The rate of ICD implants in the UK was 72 per million per year in 2010, the vast majority of which were provided to adults between 55-89 (Murgatroyd et al., 2014).

Psychological effect of ICDs- Adults

Published research indicates that adults with ICD devices may be at risk for a range of psycho-social morbidities including anxiety and depression, although prevalence rates vary widely from 8-63% for anxiety symptoms and 5-41% for depression (Magyar-Russell et al., 2011). Other concerns such as child-rearing, financial stability, interference with social interactions and sexual encounters, stress and feeling 'different' from healthy peers have been shown in adults under 50 years of age with ICDs (McDonough; 2009; Sears et al., 2001). There is general consensus in the literature that adults often experience ongoing psychological, social and physical changes and anxieties regarding unexpected electric shocks from ICDs (Camm et al., 1999; Palacios-Cena et al., 2011).

Psychological effect of ICDs-Children and Adolescents

In 2010 to 2011, one percent of ICD implants out of 3689 in England and Wales, were for patients under 20 years of age (NICOR, 2011). As this is a small population, research on adolescent adjustment to having an ICD is limited. One Australian qualitative study with adolescents and their parents identified themes related to restrictions imposed on young people: the shock experience, ongoing challenges post-implant and positive benefits of having an ICD (Rahman et al., 2012). Other quantitative research suggests that children with ICDs rate their quality of life (QoL) as comparable to other children with chronic health difficulties, with the exception of lower physical QoL (Sears et al., 2011).

Children and young people with ICD devices have to negotiate the tasks of growing up while coping with their ICD device, including the burden of ongoing medical monitoring and ICD replacement surgery. As a result, adolescents with ICDs will have to cope with more operations throughout the course of their lives than older patients. Anxieties and issues that paediatric patients with ICDs experience will vary at different developmental stages, due to the tasks of each stage and the young person's developing cognitive abilities (Piaget, 1976).

Body Image in Patients with Health Conditions

Definitions of body image describe the psychological construct as comprising thoughts, feelings, behaviours and beliefs which lead to self-perception and evaluations about one's body (Cash, 2002). Issues of body image may be particularly salient for adolescents with health conditions, as they are at higher risk

of internalising symptoms (Lavigne & Faier-Routman, 1992) and impaired development of attractive sense of self (Suris, Michaud & Viner, 2004) than healthy peers. In addition, scars can be a powerful reminder of surgery and have been shown to result in lowered self-esteem and poor body image in adult patients. (King et al., 2009). Unfortunately results are often limited by risk of bias from samples of convenience and small sample sizes, reducing the generalisability of findings to other populations.

Social acceptance and peer relationships are important factors in the development of self-esteem and body image (Karazsia, van Dulmen, Wong & Crowther, 2013). Adolescents may be particularly sensitive to this as construction of identity and integration with peer group are key tasks for their developmental stage (Erikson, 1994). For example, peer perceptions of athletic ability have been shown to correlate strongly with physical attractiveness and social acceptance in school (Vannatta, Gartstein, Zeller & Noll, 2009). ICDs limit the physical activity patients can engage in (Arrhythmia Alliance, 2008), possibly restricting the use of athletic ability as a route to achieving desirable social interactions.

The effects of having an ICD on body image and self-esteem in children and adolescents have yet to be explored in detail. These factors may be important to ensure the best psychosocial adjustment to a lifelong treatment for a chronic condition. Concerns regarding body image and self-esteem may be particularly salient where visibility of the device or scar tissue is an issue. Therefore, exploration of the anxieties and challenges of adjusting to life with an ICD may help to inform cardiac teams on how to support this group to adjust successfully post implant.

Aims

This study will use IPA to explore the effect of having an ICD device on body image and self-esteem in adolescents, and other challenges they encounter. It is hoped that this will add to the literature base regarding the effect of ICDs on this population, and may inform the development of support that is available to these patients.

Method

Ethics

Ethical approval for this project was obtained by the West of Scotland Research Ethics committee (See Appendix 2.6) and from Clinical Governance in the NHS Greater Glasgow and Clyde health board. The study was carried out in accordance with the BPS Code of Human Research Ethics (2010). The Data Protection Act (1998), NHS Data Protection and Confidentiality Policies informed the handling of data for this project. Consent procedures were informed by guidance from the Scottish Children's Research Network on obtaining informed consent (ScotCRN, 2012).

Design

This study utilised a qualitative research design comprised of in-depth individual interviews, each lasting between thirty minutes to one hour. The design was informed by the chosen method of analysis, Interpretative Phenomenological Analysis (IPA). IPA is an approach which uses semi and unstructured interviews with participants to explore their lived experiences (Smith, Flowers & Larkin, 2009).

Recruitment and Participants

Participants were recruited from the Cardiac team at the Royal Hospital for Sick Children, Glasgow between November 2014 and May 2015. The Consultant Cardiologist identified participants who met the inclusion criteria (see Table 1) and these individuals were invited to participate via letter, which included an age appropriate information sheet about the study. Three participant information sheets (PIS) were written with age appropriate language for ages 12-15 years, 16-17 years and parents. One sample PIS is included in Appendix 2.7. Sample consent and assent forms are included in Appendix 2.8. Individuals interested in participating, and who did not meet exclusion criteria, were invited to opt in by post, email or telephone.

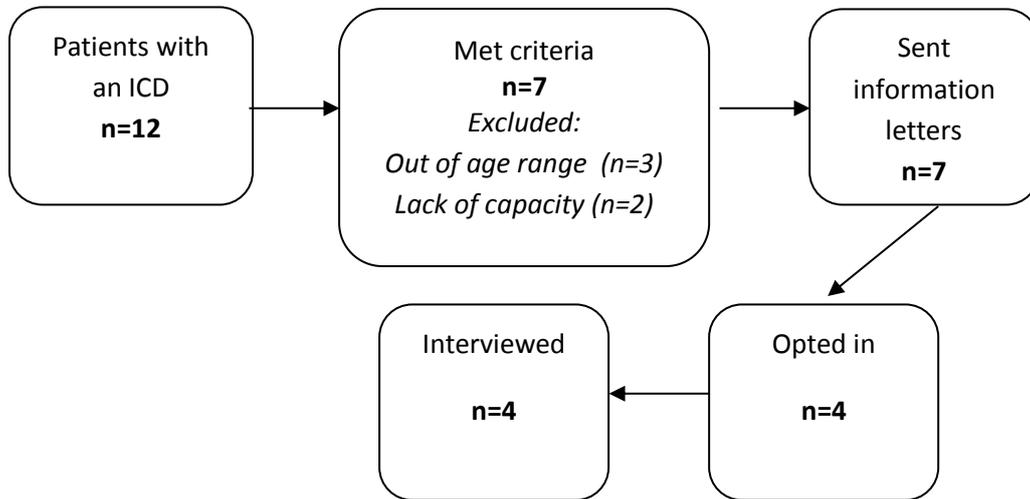
Table 1: Inclusion and Exclusion Criteria

Inclusion Criteria	Exclusion Criteria
<ul style="list-style-type: none">• Young people who have an ICD• Between the ages of 11 and 17• Duration of ICD implant minimum of six months.• Under care of RHSC, Yorkhill	<ul style="list-style-type: none">• Non English speaker• Lack of capacity to provide informed consent

Once participants had opted in, the researcher contacted them directly to co-ordinate interviews at the RHSC, Glasgow. Informed consent was obtained in written form from participants prior to the interview and written assent was also obtained from a parent (ScotCRN, 2012). At this time participants and their parents had the

opportunity to ask further questions, and confidentiality was discussed. Parents were asked to wait in a different room during the interview. A flow diagram outlining recruitment is detailed in Figure 1.

Figure 1: Flow Diagram for Recruitment



The final sample comprised of four white Scottish males, aged between 12-17 years old. Demographic information regarding the final participant sample is summarised in Table 2. Participants are identified by pseudonyms to protect anonymity. One participant had a tracheostomy tube in addition to his ICD, which may influence his experiences and perception of the ICD. His co-morbidity will be taken into consideration when discussing the findings.

Table 2: Demographic information of participants

Participant	Age at interview	Gender	Scottish Index of Multiple Deprivation Decile	Age at ICD implant
George	16	Male	5	15
Michael	12	Male	2	3
Stuart	15	Male	1	11
Joshua	17	Male	4	9

Interview Procedure

In-depth, semi-structured interviews were carried out individually with participants. The interview schedule comprised of open-ended questions aimed at drawing out discussion and reflection from the participant about their experiences with their ICD. The interview schedule was designed by the interviewer and was informed by recommendations from the literature on how to optimise participants' reflective engagement in the interview process (DiCicco-Bloom & Crabtree, 2006; Smith et al., 2009) and through discussion with the research team. Theoretical underpinnings were drawn from literature on health representations and stress and coping models (Lau & Hartman, 1983; Wallander & Varni, 1995). The interview schedule was flexible and intended as a guide, providing prompts to aid discussion where necessary. The interview schedule (see Appendix 2.9) was revisited by the researcher after two interviews to assess the suitability of the questions, and no change to the structure or questions was deemed necessary.

The interviews were carried out in a clinic room in the RHSC, Glasgow by the researcher, using a digital voice recorder (DVR). The data were subsequently transcribed verbatim and anonymised for all references to persons or places. The recordings were saved to an encrypted laptop for transcription, and once this process was complete and transcriptions were checked for accuracy, the recordings were destroyed.

Data Analysis

The data were analysed using IPA which aims to understand the experience of a particular life event for the individual and the meaning they attach to the event, in the context of their personal and social experiences (Larkin & Thompson, 2012). IPA utilises a six step procedure for data analysis, which is outlined in Table 3.

*Table 3: Procedure for Data Analysis with IPA**

Stage	Description
1: <i>Reading and re-reading</i>	Emersion in the data to promote active engagement with the participant's experiences
2: <i>Initial noting</i>	Exploratory examination of the semantic, linguistic and conceptual content of the data
3 <i>Developing emerging themes</i>	Analysis of the initial coding to identify emerging themes
4: <i>Searching for connections across emergent themes</i>	Mapping emergent themes across the text to identify the most salient and interesting concepts.
5: <i>Moving to the next case</i>	Repeating the process with each interview.
6: <i>Looking for patterns across cases</i>	Synthesising emergent themes across cases to identify super-ordinate themes

**Summary of procedures outlined in Smith et al., (2009)*

In order to minimise potential bias and to ensure the plausibility of the interpretation, a cross-section of the data were independently analysed by two research supervisors to identify common themes in the data. Supervision was also utilised throughout for this purpose, as recommended by Smith et al., (2009). A subsection of the data with IPA process notes can be found in Appendix 2.10.

Reflexivity

IPA recognises that the researcher's understanding of the participant's experience is affected by the filter of their own experience, knowledge and biases, and this will be acknowledged in the analysis and discussion of the data (Smith et al., 2009). The researcher is a thirty two year old female Trainee Clinical Psychologist who has a special interest in Child Psychology and has been working with children and families for over ten years. Consequently, the researcher used their knowledge of psychological literature pertaining to child development to understand participant experiences and to extrapolate key themes. The author does not have an ICD or any prior experience of working with individuals with these devices. Having worked in a paediatric setting, the researcher has experience providing psychological support for patients with chronic and/or severe health difficulties and co-morbid psychological distress. As a result, the author has prior knowledge of the challenges which may arise for this population.

Both academic supervisors who reviewed the transcripts are female and employed by the University of Glasgow, with a background in health psychology and clinical psychology research, and who are experienced in the use of IPA. Neither of these researchers had any contact with the participant group prior to or during the research.

Results

The four participants had varying experiences of living with their ICD and had had their devices for different lengths of time (range 1-9 years). They were able to verbalise their thoughts, although there was a notable difference in the depth of information and reflection between the older and younger participants, which may reflect different developmental stages. The two older participants were more reflective and seemed to identify more easily ways in which their ICD had influenced on their sense of self. Despite their differing developmental stages, common themes emerged which were described by at least three, if not all four, of the participants.

Six superordinate themes were identified which encapsulated the key aspects of the participants experiences (Table 4).

Table 4: Superordinate and Emergent themes

Superordinate Themes	Sub-Themes	Participant
1) Physical effect	i) Physical discomfort of ICD	1,2,4
	ii) Restriction of activity	All
	iii) Risk of harm	1,2,4
2) Emotional impact	i) Emotional distress	All
	ii) Effect on family	All
	iii) Vigilance	1,2,4
3) Sense of self	i) Social reactions	All
	ii) Visibility	All
	iii) Conflict	All
	iv) Identity	All
4) Coping Strategies	i) Knowledge	All
	ii) Avoidance	All
	iii) Carrying on	1,3,4
	iv) Supports	All
5) Development over time	i) Change	1,2,4
	ii) Overcoming challenges	1,2,4
6) Evaluation of ICD	i) Positive Outcomes	All
	ii) ICD as back-up	1,3,4

Superordinate Theme One: ‘Physical effect’

All the participants identified that their ICD device had a significant physical effect on their lives, which seemed to manifest itself in three key areas: Physical discomfort of ICD, Restriction of activity and Risk of harm.

Physical discomfort of ICD

When individuals suffer cardiac arrhythmia, the ICD uses a small electric shock to induce a regular heart rhythm. Two of the participants described the ICD shock experience as a powerful negative experience.

Have you ever touched an electric fence? D'you know the initial shock of touching an electric- you just go like that [indicates tensing up], you just all tense up and everything goes like that. It's like that, but a hundred times worse, like aw, woah.

[George, P11: 207]

“They said that if, like it shocked it would like be like a horse kicking you in the chest, it's more. And the way I was..... I actually screamed out in pain 'n that.”

[Joshua, P43: 666]

The two other participants did not think they had experienced shocks, but one described that his ICD caused him pain when it got knocked.

“Yeah it does hurt, and it makes you cry and all that.”

[Michael, 13:202]

Restriction of Activity

Relevant to all were the limitations placed on participants as a result of their ICD, which have a significant effect on their ability to engage in physical activities.

I know what I can and can't do, as I say, I can't, I shouldn't be going on, like, rides and I can't do what, I can't do contact sport, or can't jump into a swimming pool, em, I can't be next to , like, loudspeakers and stuff.

[Joshua, P8:122]

“I can’t really do, well, I haven’t done, like water skiing and stuff like that now....Yeah, so monkey bars and stuff like that, so can’t really do that.”

[George, P7: 127]

Two of the participants seemed to have accepted these limitations and integrated them into their daily activities, while the two others seemed to be struggling to reconcile these restrictions with their desired activities. This seems to reflect differences in their developmental stages, even where chronological age was similar.

“Well, it kinda does bother me, coz, like I do want to do stuff which is physical, and I know I can’t ”

[Stuart, P20: 344]

Risk

Three participants discussed events or issues which highlighted the sense of risk that they live with as a result of their heart conditions.

Obviously I am worried for my safety. I am always worried that, like, if it happens and no one is around, or that, but, as I say, hopefully someone should come along. It’s only happened a few times outside, but there has always been somebody there, usually.

[Joshua, P9: 133]

Em, my big brother’s had, like, three shocks maybe.....Yeah, his heart stopped.....: No, he hasn’t told me about it, but I know about one when he, he was running and , em, like, his body , just like, fell down to the ground and he died for seven minutes.

[Michael, P15: 225]

Superordinate Theme 2: Emotional Impact

Another main theme which was identified was the emotional impact of having an ICD. This gave rise to three main sub themes: Emotional distress, Effect on family and Vigilance.

Emotional Distress

Emotional distress for the participants included anxiety and fear, loss, frustration, pain and hopelessness. One participant described how he felt when he first received his ICD:

There was a lota, a lot of emotion I think, there was a lot of emotion showing, a lot of time you just couldn't be bothered with what the day would bring, or even when I was stuck in here quite a while, sometimes after it I just almost gave up on everything 'n it was like I couldn't be bothered with anything or I just, [pause] it's like the whole world was against me.

[Joshua, P22: 333]

The extent of distress varied between participants, with two identifying periods of significant distress and two reporting low levels of emotional upset. When asked whether he had noticed any changes in his feelings since getting his ICD, one participant commented:

"I don't know, maybe a bit of anxiety, sometimes, in certain situations. But, not, not majorly though."

[George, P8: 146]

Effect on Family

All the participants described the emotional impact of their health conditions on their family members, most commonly displayed in parental anxiety and vigilance for their children.

“Probably a bit scary, coz like there is four of us is got it, so she’s [mother] a bit scary about it.”

[Michael, P19: 293]

“Well, I think well, half the times I think my mum is a bit overprotective”

[Stuart, P3: 43]

Often this seemed to be demonstrated through monitoring of medication and reminders about limitations.

“so she was just: have you taken your pills, have you taken your pills, have you taken your pills.”

[George, P6: 110]

Vigilance

Three of the four participants described being vigilant in monitoring their own health, including self-monitoring of physical symptoms, being careful and being wary of physical contact with others as a result of their ICD.

“sometimes if your heart’s racing or whatever, well if you are upbeat or something like that , I need to watch because at the start it went off a couple

of times, three times”

[George, P2: 30]

“Yeah, it’s just like any touch. I don’t let anybody touch it basically.”

[Michael, P13: 206]

Superordinate Theme 3: Sense of Self

Issues regarding establishing a sense of self in relation to the ICD device were prevalent for all participants. The sub themes which emerged were: Social reactions, Visibility, Conflict and Identity. This superordinate theme is particularly related to the participants’ body image and worries about negative social judgements from others in relation to their ICD devices. As well as the construction of body image and self-esteem, it became apparent that integration of the ICD into sense of self had influenced their developing identities.

Social reactions

“Some people just don’t like the fact that it’s something solid underneath my skin or whatever, but, I don’t know, people think it’s a bit gross”

[George, P9: 169]

Although all had experiences of negative reactions from others, this was only a minor concern for some of the participants. For one, however, this was an ongoing concern after a particularly difficult experience in the past.

“ I went swimming with my family, and the lifeguard told me to get out, coz, like, it put off people swimming.”

[Michael, P6: 87]

All the participants had experiences as being treated differently as a result of their ICD, which either resulted in a positive sense of status or a negative sense of being singled out as different from others.

“obviously I was that young that you think ‘aw cool, I can be different from other people”

[Joshua, P3: 49]

Visibility

This theme links directly to experiences of body image for the participants. All participants identified being aware of the visibility of their ICD device, and the scarring resulting from operations. One participant did not find the visibility of his device problematic:

For a while it was a bit of a party trick when I was out with my pals, coz you can see it and feel it underneath my skin, so I used to take my top off and show everyone.

[George, P9:163]

Others were more self-conscious about the visibility of their ICDs and scars, and described trying to conceal them in public situations.

It is more to do wi’, like, the scars that have been left behind I think. As I said before, em, I think that sort ae, slightly lowers my confidence as well, say I go swimming- I am always, like that [covers chest with arms]. I know it’s stupid, but, it’s just, I probably actually should be proud of them because, but at the same time there is always that slight embarrassment. What if somebody says

something?

[Joshua, P39: 599]

I don't know. It's just like, when I go swimming I have to wear, like, wear a top and that... It's just to like, hide it because I am nervous..... Just probably, just in case if anybody makes fun of it. [Michael, P5: 72]

Conflict

Each of the participants displayed some inner conflict or dilemma regarding the ICD, although these varied between individuals. Everyone described a conflict between the two sides of the ICD as it has both benefits and drawbacks.

"I think that it is, keeps, it's there to keep, like, my heart going..... Em, well, it kinda depends on how you look at it. Because, well, there's, like, limitations to it." [Stuart, P2: 27]

It was, was mixed emotion, cos like, one moment, on one side I was like chuffed because the fact that it saved my life basically, and then on the other side was the emotional side that, what about this, what about this, the risks and stuff like that. [Joshua, P7: 103]

Salient for three of the participants was the conflict between the reality of having the ICD and the ideal life which they aspired to, which led to a sense of frustration or loss.

“I would like to be a policeman, but I don’t know if it will stop me doing that or whatever.” [Michael, P20: 321]

Like, I’d love it if I could be, like in the armed forces or, like, the RAF or kind of, like that…… well, yeah, I kinda do have to think about it more and I don’t want to have to, like, I don’t want to get a job that’s like working in an office, or anything like that. [Stuart, P12: 185]

Identity

There also seemed to be an effect on how participants integrated the ICD into their sense of identity, with two individuals seeming to have incorporated the limitations of the ICD in with their own personal qualities.

“But, I’m not, I’m really not that fussed by it, I just sit and, I’m quite a lazy person” [George, P18: 352]

“I am not allowed to do contact sport, so I’m not, I’m not really a P.E person, [laughs] so I am more into, like, your collecting or computer games now” [Joshua, P19: 273]

At the same time, there seemed to be a sense that that ICD was a separate entity which seemed intrusive to some participant’s sense of self. When asked about what he thought of the ICD device, one participant commented:

“I don’t like, really like it inside me” [Michael, P16: 252]

While another illustrated this in the way he referred to his device:

“Other than that thing there [indicates towards chest area/ ICD]”

[George, P10:191]

Superordinate Theme 4: Coping Strategies

All the participants demonstrated a variety of coping strategies which they had developed in relation to managing their ICD. The most frequently occurring strategies were: Knowledge, Avoidance, Carrying on and Supports.

Knowledge

Knowledge seemed to be a useful coping strategy for some participants and they seemed well informed about how their ICD worked. This seemed to be protective for some participants as knowledge helped them to rationalise the negative aspects of the device, promoting successful adjustment.

I went down south to Newcastle to get it put in, and they don't, up here I think they use metabolic ones, which look a bit different to the one I've got. I've got a Boston scientific one, but my one is a bit bigger.

[George, P13: 243]

In contrast, one of the younger participants displayed much less knowledge about their ICD.

“I would just say, like, it’s like a square box inside me.”

[Michael, P3: 35]

Avoidance

Avoidance was a prevalent coping strategy for all the participants as they described trying not to think or talk about their ICDs, with mixed levels of success. This seemed to enable them to continue with daily activities despite the sense of risk attached to living with their ICDs.

“No, I generally forget about it most of the time.”

[George, P8: 158]

“A lot of the times you don’t even realise that it’s there.”

[Joshua, P15: 222]

Carrying On

One of the most commonly reoccurring themes was the idea of carrying on, enduring or just having to ‘get on’ with things. This approach was apparent for three out of the four participants, and seemed like a helpful approach for coping with the day to day effect of the ICD on their lives.

“I know I’ve got all this. I just like, try to live life as good, like, in between.”

[Stuart, P9: 136]

“it does affect my life still, it always will but all of the times you just, as I say, you just get on with it.”

[Joshua, P11: 154]

Supports

Three of the participants identified supports which had helped them cope with their ICD devices, including family, peers and relationships with the medical staff in the hospital.

Talking to people has probably helped a bit, em, one of my dad's, mum and dad's friends, he's got a pacemaker in, so, like it's kind of comforting to know that someone else has kind of got the same thing as me.

[George, P16: 315]

Talking to them, you know like chatting, em, saying like, sorta hearing other situations that people have been in, I think that helped because, like It's sorta like, as if there is a feeling of hope still left, in that way.

[Joshua, P37: 558]

Superordinate Theme 5: Development over time

One striking theme that surfaced with three of the participants was the sense that their relationship to their ICD had changed over time, developing from a sometimes difficult beginning in to a stage of stability, if not acceptance. The sub themes which emerged were: Change and Overcoming challenges.

Change

“coz I am quite a forgetful person, I, I usually, well not so much anymore, but at the start I would forget it a lot”

[George, P6: 108]

“I’ve tried to stop running as much, I just, like, walk about with friends.”

[Michael, P22: 348]

These quotations illustrate some of the behaviour changes participants had made over time in relation to their ICDs. There was also a change in the effect of the ICD over time. One participant identified that there had been a gradual improvement in the extent to which the ICD influenced his life.

“It’s not really affect- if you know what I mean? Doesn’t affect as much as it did before..”

[Joshua, P16: 239]

Overcoming challenges

There was a sense of overcoming challenges throughout the time of having an ICD, with participants often describing a difficult beginning followed by a period of being settled or stable.

It took quite a long time, like for the parameters of the device took quite a while to kinda settle out, which I kind of.... expected, excuse me. Em, but that, once that, now that that’s settled it hasn’t gone off in months.

[George, P18:364]

After about a year it started to get easier. But then there would always be, find something that I couldn’t do, the it would go back and, em, but I think after the year/ two years that’s when I pretty much was, I was, like I got

used to it.

[Joshua, P21: 304]

Superordinate Theme 6: Evaluation of ICD

Overall the participants evaluated their experiences of their ICD as having a significant positive influence on their lives. The subthemes which were evident were: Positive outcomes and ICD as a back-up.

Positive Outcomes

Three of the participants evaluated the ICD in a positive light, with several significant benefits displayed throughout their interviews including a sense of increased independence and confidence. One participant reflected on the effect the ICD had had on him as a transformational experience:

Well, the good things, probably the fact that I've seen sort of, life in a different light, and I think that even the bond between my family and me is grew stronger as well. And some of the people I have met here have been just amazing, and I don't know how I could have done it without them or, and I am thinking as well, that if I hadn't of had that then I would never have met them. Em, I know it has come to an end, but at the same time I am glad it happened, I am glad it lasted while it did. And I am glad that I met them in the first place. [pause] I just became a better person because of it.

[Joshua, P49: 791]

ICD as a back-up

The majority of the participants valued the ICD as a back-up and acknowledged the role the ICD played in regulating their heart. This positive evaluation seemed to make it easier for these participants to accept the negative aspects of their devices.

“Em, I think that it is, keeps, it’s there to keep, like my heart going, so, it’s like a back-up really.”

[Stuart, P2: 22]

...It’s always looking out for me, I kind of see it like that. Em, that, it’s a kind of safety thing rather than a hindrance, so I know that, that my heart’s always, there’s not gonna be any, like, there’s not going to be any problems with my heart, coz that’s put in it kind of always looking after, checking what’s going on. Which I think it’s a comfort thing.

[George,P17: 27]

Discussion

The aim of this study was to explore the effect of having an ICD device on body image and self-esteem in adolescents, and other challenges they encounter. Overall there was a general consensus that, despite the tribulations associated with living with an ICD, the benefits of survival outweighed these. There was also an overarching sense that the effect of the ICD improved over time for the participants. Six superordinate themes emerged during the analysis: Physical effect, Emotional impact, Sense of self, Coping Strategies, Development over time and Evaluation of ICD. These themes create a useful structure for understanding young people’s

experiences of having an ICD. Although these themes have been separated out for the purposes of this study, it is useful to consider them as an integrated whole.

Most salient to the primary research question was the theme of impact of the ICD on participants' sense of self, which incorporated social reactions, visibility, conflict and identity. Social reactions are a powerful influence in the development of body image, self-esteem and identity (Karazsia et al., 2013), and are particularly important during adolescence as peer acceptance is one of the main developmental tasks (Erikson, 1994). Experiences of social reactions to participant's ICDs were mixed, with participants identifying acceptance and positive reactions from peers and family as well as negative reactions from others. For some, worries regarding negative social reactions were particularly salient, especially in public environments such as at the swimming pool. The visibility of their ICD was a particular issue in these situations, unsurprisingly, as a visible health condition puts individuals at higher risk of negative social responses such as teasing, and is widely associated with higher risk of difficulties with social interaction and negative self-perceptions (Rumsey & Harcourt, 2004).

In addition, all participants identified being treated differently by others which they viewed with mixed feelings. In some cases this led to frustration and a desire for equivocal treatment with peers which seemed to engender a feeling of disempowerment and frustration. This may be exacerbated by restrictions on physical activity. In other cases, being treated differently was also viewed positively as it conferred status on to the individual. Overall participants thought that there were no significant differences in functioning compared with peers, other than in the

domain of physical exercise. This finding is consistent with the literature as children and adolescents with ICD devices have been shown to appraise their QoL functioning as comparable with healthy peers with the exception of physical quality of life (Sears et al., 2011).

Central to the issue of body image were the visibility of the ICD and scars. All participants were conscious of the visibility of their implant, although reactions to this varied. The literature suggests that while visibility of a health condition puts individuals at higher risk of low self-esteem and poor body image, this is likely to be affected by factors such as prior experience of negative social reactions (Rumsey & Harcourt, 2004). In this study, George had less experience of negative social reactions and appeared to have less body image concerns than the other three, which may be expected as self-evaluation of one's body is influenced by social and cultural factors (Fan & Eiser, 2009). One participant also had a tracheostomy tube which was more prominent than his ICD, making it difficult for him to separate out the influence of his implant on body image. Although he chose not to discuss issues regarding body image explicitly, he did identify that he would not get changed in public situations, indicating some underlying awareness of this. The visibility of the ICD and resulting scars may be a risk factor for body image issues (Gabriel & Danilowicz, 1978; King et al., 2009), however this does not seem to be the case for all of the participants.

An interesting finding in this study was the emergence of themes of conflict and identity which were present for all participants. Conflict between the reality of having an ICD and the 'ideal', such as being able to pursue a particular career,

seemed linked to the negative evaluation of aspects of the ICD. This issue commonly arises for adolescents without health problems as well, e.g. with limitations arising from poor academic results. This is likely to be linked to the developmental stage of participants, as older participants seemed to have resolved these dilemmas more successfully. Similarly, the older adolescents seemed to have accommodated the ICD into their identity, cognitively reframing the limitations as fitting in with their personalities, likes and dislikes, a process common to developmental models of cognition (Piaget, 1976). This developmental process may be different in adults and older adults with ICDs, as their sense of identity is likely to be more established and the ICD may be less integrated with aspects of their personalities. Research into this area would be required to explore this theory. In addition, a development over time was identified in the data, where participants altered their behaviour in relation to the ICD, and where they perceived the ICD to 'settle in' after an initially difficult period. This suggests that most of the participants were able to adjust successfully to their device over time, similar to outcomes for other chronic health conditions (Eiser, 1990).

A number of challenges to living with an ICD were identified, including the aversive physical experience, emotional impact and restrictions on activity, which are consistent with previous quantitative and qualitative findings on paediatric adjustment to ICDs (Rahman et al., 2012; Sears et al., 2009). The negative outcomes of having an ICD were tempered by the overall positive appraisal of the device as a life-saving machine and the unanimous agreement from participants that they had to simply 'carry on' in order to overcome challenges. In order to achieve a lifestyle similar to their peers, participants adopted a number of coping strategies including

knowledge, avoidance and reliance on external supports. The literature on adjustment to chronic health suggests that knowledge is a key coping mechanism in reduction anxiety and increasing adherence to treatment (Beeri, Haramati, Rein & Nir, 2001), and the use of adaptive coping strategies has been identified as a significant protective factor for child adjustment to their health condition (Wallander & Varni, 1995). Parental anxiety was identified as a theme for all participants and is consistent with the evidence base for psychosocial effect on parents for having a child with congenital heart disease (Lawoko, 2007).

Finally, it is important to acknowledge the differences between the developmental stages of participants and how this affected their ability to verbalise their experiences. Two participants were more concrete in their thinking and seemed to find it harder to reflect on their emotional and cognitive experiences. One of them had his ICD since pre-school age, in comparison to the rest who were slightly older at the time of implant. This may have made it difficult for him to identify changes which occurred as a result of the ICD as he had no memory of living without it. In addition, his lack of knowledge about the device may also be a consequence of his age at implant, as he would have been unable to seek information himself and possibly less likely to retain it over time.

Strengths and Limitations

This study has a number of strengths which add validity to the findings. Despite the small sample size, four participants represent over half of the eligible sample and a third of the overall population of young people under 18 with an ICD who attended the RHSC, Glasgow at the at the time of the study. As the RHSC is the main centre

for ICD implant in Scotland, this figure is likely to be representative of the population across the country. In addition, the homogeneity of the sample is a strength, as are individual interviews which allowed in depth exploration of the topic. The length of recruit time maximised the likely response rate from the population.

In terms of limitations, there were variations in the developmental stage and expressive ability of the participants which may have affected the quality of the responses in some cases. Having all male respondents reflects the fact that there were fewer females with ICDs than males attending the RHSC. This is representative of the gender differences in the ICD population in England and Wales as there are fewer women than men with the implant across the lifespan (3073 men compared with 615 women in 2010-2011, (NICOR, 2011)). The findings may not represent the experiences of females with ICDs, particularly in relation to body image. In the general population, females are more likely than males to experience negative body image and place greater investment in their appearance (Muth & Cash, 2006), therefore may be more susceptible to body image difficulties as a result of their ICDs. The small sample size also means that data saturation may not have been reached, although all the superordinate themes were present for at least three, if not all, participants. This suggests that the themes which did emerge were salient for the population. Finally, one of the participants also had a tracheostomy tube which is likely to have affected his experience of the ICD device and his adaptation to his chronic health conditions in general.

Implications for Clinical Practice

The findings of this study contribute to the growing research base regarding experiences of children and adolescents with ICDs. The findings provide an insight into some of the key themes which arise for this population, which can inform future practice and service delivery for paediatric cardiac teams working with adolescents with ICDs. Promoting positive physical and emotional coping strategies such as identifying personal strengths are likely to facilitate good adjustment to having an ICD. Social networks are associated with improved health related QoL for paediatric patients with pacemakers (Cheng et al., 2014), therefore opportunities to increase social supports, including interaction with peers with similar difficulties, are likely to have positive outcomes for patients. Peer supports may provide a valuable source of information, normalisation and an opportunity to establish a protective peer group. This may have benefits for the establishment of identity as well as body image and self-esteem. Cardiac services could link into other services to facilitate activities or educational groups for this purpose. Paediatric services often utilise a developmental perspective to support how they interact with children and young people, tailoring information to different age groups. An emphasis on this approach may enable them to recognise changes over time and address the ambivalence which patients may experience regarding the positive and negative aspects of the ICD.

Implications for Future Research

A larger, multicentre qualitative study would be useful to build on the findings of this study. Including females would also be useful to understand whether their experiences of their ICDs are similar to males, or whether different themes emerge. Future research into how children and adolescents' developing identity is affected by

ICD implant at different ages would also be useful to understand the long term implications of having an ICD from a young age, and to identify the cognitive processes which result in positive adjustment to the ICD over time. Comparisons with adults with ICDs may also inform medical teams whether there are differences in the processes underlying successful adjustment for different age groups, and how best to support these at each developmental stage.

Conclusions

The physical and emotional impact of the ICD emerged as significant themes for participants, with ICD influencing participants' sense of self, including aspects of body image and identity. Although participants identified a number of negative consequences of having an ICD there was an over-arching positive evaluation of the device, a determination to overcome challenges and a positive progression in adjustment over time.

References

Arrhythmia Alliance (2008). *Physical activity and exercise advice for patients with an ICD*. Retrieved from <http://www.heartrhythmcharity.org.uk/>

Bardy, G., Lee, K., Mark, D., Poole, J., Packer, D., Boineau, R.,... & Ip, J., (2005). Amiodarone or an implantable cardioverter-defibrillator for congestive heart failure. *The New England Journal of Medicine*, 352(3), 225-237.
doi: 10.1056/NEJMoa043399

Beeri, M., Haramati, Z., Rein, J. J., & Nir, A. (2001). Parental knowledge and views of pediatric congenital heart disease. *The Israel Medical Association Journal: IMAJ*, 3(3), 194.

British Psychological Society (2010). *Code of Human Research Ethics*.
Leicester: The British Psychological Society

Camm, A. J., Sears, S. F., Todaro, J. F., Lewis, T. S., Sotile, W., & Conti, J. B. (1999). Examining the psychosocial impact of implantable cardioverter defibrillators: a literature review. *Clinical cardiology*, 22(7), 481-489.
doi: 10.1002/clc.4960220709

Cash, T. F. (2002). Cognitive-behavioral perspectives on body image. In Cash & Pruzinsky (Eds). *Body image: A handbook of theory, research, and clinical practice*, (pp. 38-46). New York: Guilford Press

Cheng, P., Gutierrez-Colina, A. M., Loiselle, K. A., Strieper, M., Frias, P., Gooden, K., & Blount, R. L. (2014). Health related quality of life and social support in pediatric patients with pacemakers. *Journal of Clinical Psychology in Medical Settings, 21(1)*, 92-102. doi: 10.1007/s10880-013-9381-0

Data Protection Act 1998 (1998). London: Stationery Office

Di Cicco-Bloom, B., & Crabtree, B. F. (2006). The qualitative research interview. *Medical Education, 40(4)*, 314-321. doi: 10.1111/j.1365-2929.2006.02418.x

Eiser, C. (1990). Psychological effects of chronic disease. *Journal of Child Psychology and Psychiatry, 31(1)*, 85-98.
doi: 10.1111/j.1469-7610.1990.tb02274.x

Erikson, E. H. (1994). *Identity: Youth and crisis* (No. 7). New York: WW Norton & Company.

Fan, S., & Eiser, C., (2009). Body image of children and adolescents with cancer: A systematic review. *Body Image, 6*, 247-256.
doi:10.1016/j.bodyim.2009.06.002

Gabriel, H. P., & Danilowicz, D. (1978). Postoperative responses in 'prepared' child after cardiac surgery. *British Heart Journal, 40(9)*, 1046-1051.
doi:10.1136/hrt.40.9.1046

- Karazsia, B. T., van Dulmen, M. H., Wong, K., & Crowther, J. H. (2013). Thinking meta-theoretically about the role of internalization in the development of body dissatisfaction and body change behaviors. *Body image, 10(4)*, 433-441. doi:10.1016/j.bodyim.2013.06.005
- King, K., McFetridge-Durdle, J., LeBlanc, P., Anzarut, A & Tsuyuki, R. (2009). A descriptive examination of the impact of sternal scar formation in women. *European Journal of Cardiovascular Nursing, 8*, 112-118. doi: 10.1016/j.ejcnurse.2008.08.001
- Larkin, M., & Thompson, A., (2012). Interpretative phenomenological analysis. In A Thompson, A., & Harper, D. (Eds). *Qualitative research methods in mental health and psychotherapy: a guide for students and practitioners* (pp 99-116). Oxford: John Wiley.
- Lau, R. R., & Hartman, K. A. (1983). Common sense representations of common illnesses. *Health Psychology, 2(2)*, 167. doi: 10.1037/0278-6133.2.2.167
- Lavigne, J. V., & Faier-Routman, J. (1992). Psychological adjustment to pediatric physical disorders: A meta-analytic review. *Journal of Pediatric Psychology, 17(2)*, 133-157. doi: 10.1093/jpepsy/17.2.133
- Lawoko, S. (2007). Factors influencing satisfaction and well - being among parents of congenital heart disease children: development of a conceptual model

based on the literature review. *Scandinavian Journal of Caring Sciences*,
21(1), 106-117. doi: 10.1111/j.1471-6712.2007.00444.x

Magyar-Russell, G., Thombs, B., Cai, J., Baveja, T., Kuhl, E., Singh, P., ... &
Ziegelstein, R. (2011). The prevalence of anxiety and depression in adults
with implantable cardioverter defibrillators: A systematic review. *Journal of
Psychosomatic Research*, 71, 223-231.
doi:10.1016/j.jpsychores.2011.02.014

McDonough, A (2009). The experiences and concerns of young adults (18-40 years)
Living with an implanted cardioverter defibrillator (ICD). *European Journal
of Cardiovascular Nursing*, 8, 274- 280. doi: 10.1016/j.ejcnurse.2009.03.002

Murgatroyd, F., Linker, N., Cunningham, D., Cunningham, M. , Chadburn, L .,
Gilbert, S., de Lange, A., (2014). *National Audit of Cardiac Rhythm
Management Devices*. National Institute for Cardiovascular Outcomes
Research (NICOR). London: University College London.

Muth, J. L., & Cash, T. F. (1997). Body - Image Attitudes: What Difference Does
Gender Make? *Journal of Applied Social Psychology*, 27(16), 1438-
1452. doi: 10.1111/j.1559-1816.1997.tb01607.x

NHS Greater Glasgow And Clyde. *NHS Data Protection policy*.
Glasgow: NHS GG&C

NHS Greater Glasgow And Clyde . *Confidentiality policy*.

Glasgow: NHS GG&C.

National Institute for Cardiovascular Outcomes Research (NICOR) (2011).

Retrieved from <http://www.ucl.ac.uk/nicor> [Last accessed 10th July 2015.]

National Institute for Health and Clinical Excellence (Jan 2006) Implantable

cardioverter defibrillators for arrhythmias. *Review of Technology Appraisal*

11. London: NICE. Retrieved from www.nice.org.uk

Palacios-Cena, D., Losa-Iglesias, M., Alvarez-Lopez, C., Cachon-Perez, M., Reyes,

R., Salvadores-Fuentes, P. & Fernandes-de-las-Penas, C, (2011). Patients,

intimate partners and family experiences of implantable cardioverter

defibrillators: qualitative systematic review. *Journal of Advanced Nursing*,

67 (12), 2537-2550. doi: 10.1111/j.1365-2648.2011.05694.x

Piaget, J. (1976). Piaget's theory. In Inhelder, B., Chipman, H., & Zwingmann, C.,

(Eds) *Piaget and His School* (pp 11-23). New York: Springer –Verlag

New York Inc.

Rahman, B., Macciocca, M., Sahhar, M., Kameri, S., Connell, V., & Duncan, R.

(2012). Adolescents with implantable cardioverter defibrillators: A patient

and parent perspective. *Pacing and Clinical Electrophysiology*, 35(1), 62- 72

. doi:10.1111/j.1540-8159.2011.03229.x

Rumsey, N., & Harcourt, D., (2004). Body image and disfigurement: issues and interventions. *Body Image, 1*(1), 83-97. doi:10.1016/S1740-1445(03)00005-6

Scottish Children's Research Network (2012). *ScotCRN Guidance: Obtaining informed consent for clinical research in children and young people under 16 in Scotland. 7.08.12 (Version 1)*. Retrieved from <http://www.scotcrn.org/informed-consent/>

Sears, S. F., Amant, J. B., & Zeigler, V. (2009). Psychosocial considerations for children and young adolescents with implantable cardioverter defibrillators: an update. *Pacing and Clinical Electrophysiology, 32*(s2), S80-S82. doi: 10.1111/j.1540-8159.2009.02391.x

Sears, SF., Burns, J.L., Handberg, E., Sotile, W.M., Conti, J.B., (2001). Young at heart: Understanding the unique psychosocial adjustment of younger implantable cardioverter defibrillator recipients. *Pacing and Clinical Electrophysiology, 24*, 1113-1117. doi: 10.1046/j.1460-9592.2001.01113.x

Sears, S., Hazelton, G., St. Amant, J., Matchett, M., Kovacs, A., Vazquez, L., & Bryant, R., (2011). Quality of life in paediatric patients with implantable cardioverter defibrillators. *American Journal of Cardiology, 107*, 1023–1027. doi:10.1016/j.amjcard.2010.11.027

Smith, J., Flowers, P., & Larkin, M., (2009) *Interpretative Phenomenological Analysis: Theory, Method and Research*. London: SAGE Publications Ltd.

- Stefanelli, C. B., Bradley, D. J., Leroy, S., Dick, M., Serwer, G. A., & Fischbach, P. S., (2002). Implantable cardioverter defibrillator therapy for life-threatening arrhythmias in young patients. *Journal of Interventional Cardiac Electrophysiology*, 6(3), 235-244. doi: 10.1023/A:1019509803992
- Suris, J. C., Michaud, P. A., & Viner, R. (2004). The adolescent with a chronic condition. Part I: developmental issues. *Archives of disease in childhood*, 89(10), 938-942. doi:10.1136/adc.2003.045369
- Vannatta, K., Gartstein, M. A., Zeller, M., & Noll, R. B. (2009). Peer acceptance and social behavior during childhood and adolescence: How important are appearance, athleticism, and academic competence? *International Journal of Behavioral Development*, 33(4), 303-311. doi:10.1177/0165025408101275
- Wallander, J. L., & Varni, J. W. (1995). Appraisal, coping, and adjustment in adolescents with a physical disability. In J.L Wallander & L.J. Siegel (Eds) *Adolescent health problems: Behavioral perspectives*. New York: Guilford Press

CHAPTER THREE: REFLECTIVE ACCOUNT

Formulating Therapeutic Resistance

Louisa Casselden, MA (Hons), MSc

Institute of Health & Wellbeing

College of Medical, Veterinary and Wellbeing

University of Glasgow

Word count: 5221

July 2015

Abstract

Introduction. The topic for this reflective account is introduced as formulating therapeutic resistance, illustrated through the discussion of three cases with which I worked during my clinical training. This reflection is structured using Gibbs' Reflective Cycle Model (1988), which is described.

Reflection. The initial thoughts and feelings elicited by each case are explored, as are the problems encountered during the therapeutic work. My management of the therapeutic resistance is analysed, and changes in my understanding and approach to the management of this issue is analysed through discussion of the three cases. I endeavour to show the development in my formulation skills and increased understanding of the client's problems as my ability to integrate different models has improved. I discuss what I would do differently with each case now, as well as the importance of supervision and wider reading to aid reflection and improve my management of therapeutic resistance.

Reflective Review. I reflect on the process of writing the reflective account and the use of the Gibbs' model to facilitate my reflections.

CHAPTER FOUR: REFLECTIVE ACCOUNT

Working Across Services: A Reflection on Service Issues

Louisa Casselden, MA (Hons), MSc

Institute of Health & Wellbeing

College of Medical, Veterinary and Wellbeing

University of Glasgow

Word count: 5515

July 2015

Abstract

Introduction. This reflective account explores my experience of team meetings in three different teams, two in a community mental health setting and one tertiary care service. I reflect on the differences and discuss how services issues, such as structure and service re-design, may have impacted on multidisciplinary working in the teams and the atmosphere in team meetings. I used Rolfe, Freshwater and Jasper (2001)'s reflective model to structure this account, which is described.

Reflection. My observations about the structure, atmosphere and multidisciplinary working in the team meetings are explored. I then reflect on the underlying processes and wider service issues which may have been impacting on each of the teams, and how these processes were visible in team meetings. I also explore my own developmental stage as a Clinical Psychology Trainee and how this may have impacted on my experience of team meetings.

Reflective Review. I discuss the process of writing this account and my experience of using Rolfe et al. (2001)'s model to structure my reflections, with conclusions concerning future reflective practice techniques.

Appendix 1.1- Author Publication Guidelines

British Journal of Health Psychology

Author Guidelines

The aim of the British Journal of Health Psychology is to provide a forum for high quality research relating to health and illness. The scope of the journal includes all areas of health psychology as outlined in the Journal [Overview](#).

The types of paper invited are:

- papers reporting original empirical investigations, using either quantitative or qualitative methods;
- theoretical papers which may be analyses or commentaries on established theories in health psychology, or presentations of theoretical innovations;
- review papers, which should aim to provide systematic overviews, evaluations and interpretations of research in a given field of health psychology; and
- methodological papers dealing with methodological issues of particular relevance to health psychology.

1. Circulation

The circulation of the Journal is worldwide. Papers are invited and encouraged from authors throughout the world.

2. Length

Papers should normally be no more than 5000 words (excluding the abstract, reference list, tables and figures), although the Editor retains discretion to publish papers beyond this length in cases where the clear and concise expression of the scientific content requires greater length.

3. Editorial policy

The Journal receives a large volume of papers to review each year, and in order to make the process as efficient as possible for authors and editors alike, all papers are initially examined by the Editors to ascertain whether the article is suitable for full peer review. In order to qualify for full review, papers must meet the following criteria:

- the content of the paper falls within the scope of the Journal
- the methods and/or sample size are appropriate for the questions being addressed
- research with student populations is appropriately justified
- the word count is within the stated limit for the Journal (i.e. 5000 words)

4. Submission and reviewing

All manuscripts must be submitted via [Editorial Manager](#). You may like to use the [Submission Checklist](#) to help you prepare your manuscript. Before submitting, please read the [terms and conditions of submission](#) and the [declaration of competing interests](#).

5. Manuscript requirements

- Contributions must be typed in double spacing with wide margins. All sheets must be numbered.
- Manuscripts should be preceded by a title page which includes a full list of authors and their affiliations, as well as the corresponding author's contact details. A template can be downloaded from [here](#).
- For articles containing original scientific research, a structured abstract of up to 250 words should be included with the headings: Objectives, Design, Methods, Results, Conclusions. Review articles should use these headings: Purpose, Methods, Results, Conclusions.
- For reference citations, please use APA style. Particular care should be taken to ensure that references are accurate and complete. Give all journal titles in full and provide doi numbers where possible for journal articles
- In normal circumstances, effect size should be incorporated.

Full details are available at:

[http://onlinelibrary.wiley.com/journal/10.1111/\(ISSN\)2044-8287/homepage/ForAuthors.html](http://onlinelibrary.wiley.com/journal/10.1111/(ISSN)2044-8287/homepage/ForAuthors.html) (last accessed on 28th May 2015)

Appendix 1.2: Search Terms

EMBASE

1	anxiety/ or "quality of life"/ or exp psychological aspect/or psycholog*/ or psychological well being/.mp.
2	psychiatric treatment/ or "psychological and psychiatric procedures"/ or psychiatric diagnosis/
3	affect/ or parental behavior/ or psychopathology.mp.
4	(adjustment or adaptat* or coping or mood disorder).mp. [mp=title, abstract, subject headings, heading word, drug trade name, original title, device manufacturer, drug manufacturer, device trade name, keyword]
5	(mental adj2 (stress or health or disease)).mp. [mp=title, abstract, subject headings, heading word, drug trade name, original title, device manufacturer, drug manufacturer, device trade name, keyword]
6	1 or 2 or 3 or 4 or 5
7	((cardiac or cardio* or heart) adj2 (arrhythmi* or disease*)). mp. [mp=title, abstract, subject headings, heading word, drug trade name, original title, device manufacturer, drug manufacturer, device trade name, keyword]
8	implantable cardioverter defibrillator/ or artificial heart pacemaker/ or defibrillator/
9	7 or 8
10	(child* or adolescent* or p*ediatric or youth or juvenile).mp. [mp=title, abstract, subject headings, heading word, drug trade name, original title, device manufacturer, drug manufacturer, device trade name, keyword]
11	((famil* adj3 relation*) or parent*).mp. [mp=title, abstract, subject headings, heading word, drug trade name, original title, device manufacturer, drug manufacturer, device trade name, keyword]
12	6 and 9 and 10 and 11
13	Limit 12 to (human and english language and journal and child <unspecified age>)

Searched 3rd January 2015

Appendix 1.3: Quality Appraisal Tool with scoring criteria

Quality Assessment Tool – scoring criteria

Adapted from: SIGN checklist 3: cohort studies (Sleith, C., 2012) & Crowe Critical Appraisal Tool, 2013.

Psychosocial adaptation in parents with children with cardiac problems: A Systematic Review		
Authors		
Title of Article		
Title of Journal		
Date of Publication		
Completed by		
Completed on		
1	PRELIMARIES	
1.1	Does the title include study aims and design? 1 point for each	2 Well covered 1 Adequately covered 0 Poorly/Not covered
1.2	Does the abstract provide a structured summary which is balanced and informative? 2pt: yes 1pt: yes but missing some important info 0pt: no	2 Well covered 1 Adequately covered 0 Poorly/Not covered
2.	INTRODUCTION	
2.1	Scientific background and explanation of rationale 1 pt for each	2 Well covered 1 Adequately covered 0 Poorly/Not covered
2.2	Clearly defined primary hypotheses or aims (and secondary, where appropriate) 2pt: yes 0pt: no	2 Well covered 1 Adequately covered 0 Poorly/Not covered
3.	DESIGN	
3.1	Is the design congruent with the background, aims and outcomes of the research? 2pt: Design defined & appropriate 1pt: Design appropriate but not clearly defined 0pt: Design not appropriate & not defined	2 Well covered 1 Adequately covered 0 Poorly/Not covered
3.2	Are confounding variables identified and accounted for in the design and analysis? <i>E.g. Factors related to non-specific chronic illness, variable social & family circumstances</i> 2pt: Variables identified and design modified	2 Well covered 1 Adequately covered 0 Poorly/Not covered

	<p>to address them 1pt: potential confounding variables identified and discussed 0pt: Confounding variables not discussed</p>	
3.3	<p>Is the risk of bias addressed (randomised/blinded)? <i>E.g. attrition, interviewer, selection, balanced groups, equivalent treatment of participants/groups.</i> 2pt: Risk of bias identified and addressed in design/analysis 1pt: Risks of bias are identified and discussed in relation to findings 0pt: Not identified or discussed</p>	<p>2 Well covered 1 Adequately covered 0 Poorly/Not covered</p>
4.	SAMPLING	
4.1	<p>Is the sample size given? 2pt: Sample size clearly detailed 1pt: Given but not clear 0pt: No sample size given</p>	<p>2 Well covered 1 Adequately covered 0 Poorly/Not covered</p>
4.2	<p>Are details of how sample size was determined provided? 2pt: Power calculation given or discussed in relation to sample size and adequate 1pt: Power calculation discussed but not adequate 0pt: not discussed</p>	<p>2 Well covered 1 Adequately covered 0 Poorly/Not covered</p>
4.3	<p>Are details of participant characteristics given and are they representative of the population? 2pt: Details given and representative 1pt: Details given and not representative 0pt: Details not given</p>	<p>2 Well covered 1 Adequately covered 0 Poorly/Not covered</p>
4.4	<p>Are there clear inclusion & exclusion criteria. 2pt: Details of criteria clearly laid out 0pt: Details not given</p>	<p>2 Well covered 1 Adequately covered 0 Poorly/Not covered</p>
5	DATA COLLECTION	
5.1	<p>Data collection protocol reported 2pt: Enough to recreate the study 1pt: Some detail, not enough to reproduce 0pt: No details given</p>	<p>2 Well covered 1 Adequately covered 0 Poorly/Not covered</p>
5.2	<p>Method to ensure quality of measurement tool? <i>E.g. validity/reliability, pilot study, standardised tests, independent/multiple measurement.</i> 2pt: Standardised measures used and reliability and validity described for population. 1pt: Standardised tools used, no discussion of Reliability/validity OR non standardised tools used with pilot study/ analysis to explore reliability. 0pt: Non standardised tools used, no discussion of reliability/validity</p>	<p>2 Well covered 1 Adequately covered 0 Poorly/Not covered</p>

5.3	How is non- participation managed (participants lost to follow up/incomplete data)? 2pt: Non participation discussed and analysis of attrition/ non participation carried out. Incomplete data managed appropriately. Opt: No attrition analysis/ missing data not accounted for.	2 Well covered 1 Adequately covered 0 Poorly/Not covered
6.	ETHICAL ISSUES	
6.1	Participant ethical issues addressed (e.g informed consent, confidentiality, equity) 2pt: consent process identified Opt: consent process not identified	2 Well covered 1 Adequately covered 0 Poorly/Not covered
6.2	Researcher ethical issues addressed (e.g: Ethical approval, funding, conflicts of interest identified)? 2pt: Ethical approval stated/ considered in design of study Opt: Not addressed in write up	2 Well covered 1 Adequately covered 0 Poorly/Not covered
7.	ANALYSIS	
7.1	Were the statistical tests used to assess the main outcomes appropriate? 2pt: Yes 1pt: Appropriate, but other tests may have been more suitable Opt: No	2 Well covered 1 Adequately covered 0 Poorly/Not covered
7.2	Does the study identify participant flow at each stage? 2pt: Yes Opt: No	2 Well covered 1 Adequately covered 0 Poorly/Not covered
7.3	Are the results clearly reported? 2pt: Clearly reported with supporting tables 1pt: Results reported but not clearly laid out Opt: Results not reported adequately	2 Well covered 1 Adequately covered 0 Poorly/Not covered
7.4	Are confidence intervals, effect sizes and p-values reported where appropriate? 2pt: All of the above reported 1pt: P values only Opt: Not reported	2 Well covered 1 Adequately covered 0 Poorly/Not covered
8.	DISCUSSION	
8.1	Are the limitations of the study described? 2pt: Well covered 1pt: Some limitations only Opt: Not attempted	2 Well covered 1 Adequately covered 0 Poorly/Not covered
8.2	Are the results interpreted in the context of current evidence? 2pt: yes Opt:No	2 Well covered 1 Adequately covered 0 Poorly/Not covered
8.4	Is the generalizability of the research discussed? 2pt: Yes, well covered 1pt: Limited discussion	2 Well covered 1 Adequately covered 0 Poorly/Not covered

	0pt: Not discussed/ incorrect assumptions	
8.5	Do the authors provide recommendations of clinical practice or future research in relation to the findings? 2pt: yes, well covered 1pt: adequately covered 0pt: not covered	2 Well covered 1 Adequately covered 0 Poorly/Not covered
	GENERAL NOTES	
	Total score (out of 50): Percentage (%):	

Appendix 1.4 Additional demographic information from papers

Study	Parent education level	Child cardiac condition
Berant, Mikulincer & Shaver, (2008)	19.5 (31%) completed high school 39 (62%) other professional programme	Ventricular septal defects (VSD), arterial septal defect (ASD) & transposition of the great arteries (TGA)
Brosig et al., (2007)	Not reported	hypoplastic left heart syndrome (HLHS) & TGA
Doherty et al., (2009)	Not reported	Acyanotic, Cyanoitic- including Tetralogy of Fallot (TF), TGA, HLHS, Other
Franck et al., (2010)	Not reported	Open heart surgery. Conditions not specified.
Helfricht et al., (2008)	Not reported	ASD, VSD, Complex single ventricle, acyanotic defects
Hendriks et al., (2005)	low=11 middle=8 high=5	Long QT syndrome
Landolt et al., (2014)	years of education: M=14.73, SD=2.55	Non-cyanotic CHD Cyanotic CHD Other CHD
Lawoko & Soares, (2006)	Mandatory: M=43 College: M=299 University: M=237 Other: M=40	Patent ductus arteriosus, ASD, VSD, TF, pulmonary stenosis (PS), aortic stenosis (AS), TGA, Other
Majnemar, et al., (2006)	Not reported	Cyanotic lesions, TF, TGA, Septal defects, Other.
Menahem, et al., (2008)	Attended secondary=23 Completed secondary=20 Attended tertiary=1 Completed tertiary=11	VSD, ASD, Other
Solberg, et al., (2012)	years of education: Mild CHD: M=14.5, SD=2.6 Mod CHD:M=14.8, SD=2.3 Severe CHD:M=14.7, SD=2.8	HLHS, TF, TGA, VSD, ASD, Severe PS, Severe AS, Coarctation of the aorta. Other
Spijkerboer et al., (2007)	Not reported	ASD, VSD, TGA, PS
Torowicz, D., et al.,	Not reported	Single Ventricular Physiology

(2010)		Bi-Ventricular Physiology
Vrijmoet-Wiersma et al., (2009)	Low (O level)=35 Middle (A level/vocational)=63 Higher (University/ higher professional education)=95	VSD, TGA, HLHS, TF, Common Arterial trunk, AS, PS, Other
Walker, Gauvreau, & Jenkins, (2004)	Secondary=69 Vocational/college=69 College=108 Graduate/Professional=55 Not reported=20	Innocent murmur, chest pain, chronic fatigue syndrome, supraventricular tachycardia, trivia/non trivial structure disease, minor/moderate/ major interventions
Wray, & Sensky, (2004)	Not reported	Cyanotic lesions (n=29) Acyanotic lesions n=46)

Appendix 1.5: Additional Effect Sizes for Articles

Study	Effect Sizes
Berant, Mikulincer, & Shaver, (2008)	Not able to compute
Brosig et al., (2007)	$d= 0.80-1.19$
Doherty et al., (2009)	$d= 0.37$
Franck et al., (2010)	Not able to compute
Helfricht et al., (2008)	$d=0.56, d=0.14$
Hendriks et al., (2005)	$d=0.57-0.59$
Landolt et al., (2014)	$d= 0.17-0.28$
Lawoko & Soares, (2006)	$d=1-1.75$
Majnemar, et al., (2006)	Not able to compute
Menahem, et al., (2008)	$d=0.73-1.34$
Solberg, et al., (2012)	$d=0.37-0.65$
Spijkerboer et al., (2007)	Not able to compute
Torowicz, D., et al., (2010)	$d=2.43, d=0.54, d=0.2$
Vrijmoet-Wiersma et al., (2009)	Mothers: $d=0.1- 1.31$ Fathers: $d=0.16-0.82$
Walker, Gauvreau, & Jenkins, (2004)	Not able to compute
Wray, & Sensky, (2004)	Marital satisfaction: $d=0.22, 0.23$ Happiness: $d= 0.19, 0.52$ Not able to compute effect sizes for GHQ

Appendix 2.1: Author Publication Guidelines

Author Guidelines

Submission to *International Journal of Qualitative Studies on Health and Well-Being (QHW)* is taken to imply that the same manuscript is not under consideration by another journal. If the manuscript forms part of a book currently in press, the authors should specify details of the publisher and expected date of publication.

Please note that the submitting author will be the principal contact for editorial correspondence, throughout the peer review and proofreading process, if applicable.

Covering letter - in his/her covering letter, the corresponding author should reveal whether the submitted article – or very similar work - has been previously published, or orally presented, or is under consideration elsewhere.

Title page Organize the title page in the following way: 1) title of manuscript, 2) name of author(s), 3) name of department(s) and institution(s), and 4) name and full postal address of the corresponding author who also acts as 'Guarantor' for all parts of the paper. The title page should be uploaded as a separate page, and not included in the main document that will be sent out for peer review.

Language All articles should be written in English - British or American as long as consistency is observed. SI units should be used. Please subject the manuscript to professional language editing before submitting the final version if you are not a native speaker.

Acknowledgements All contributors who do not meet the criteria for authorship should be listed in an acknowledgments section. Examples of those who might be acknowledged include a person who provided purely technical help, writing assistance, or a department chairperson who provided only general support. Financial and material support should also be acknowledged.

Conflict of interest and funding Authors are responsible for recognizing and disclosing financial and other conflicts of interest that might affect their work. State relevant financial (e.g. patent or stock ownership, consultancies, speaker' fees), personal, political, intellectual or religious interests. Funding for any type of publication, for example by a commercial company, charity or government department, should be stated. This applies to all types of papers (including, for example, research papers, review papers, letters, editorials and commentaries). A conflict of interest should not prevent someone from being listed as an author if they qualify for authorship.

Title The title should be informative and accurate and at the same time trigger the interest of the reader. A short running head will be derived from the title to appear on each page of the paper.

Abstract Articles must include an abstract of up to 300 words. The abstract should stand alone, enabling a reader to decide whether or not to proceed to the full text of the article.

Keywords After the abstract, please give 5-10 key words for readers looking for material by key word searching on Internet. Avoid using the same words as in the title.

Biographical details Include full name(s), current professional affiliation, and an email address for correspondence. If more than one author, please indicate who the corresponding author is.

Section headings Please do not number section headings. Use a maximum of three levels of headings made clear by orthographic indicators, i.e. capitals, italics, bold etc.

Quotations Please use double quotation marks. Quotations longer than 40 words should appear in a separate paragraph, indented by tapping a ca 1cm right margin, without quotation marks.

Citation and Reference system - *QHW* applies the APA system. Check for full details [here](#)

Full details are available at:

<http://www.ijqhw.net/index.php/qhw/pages/view/guidelines> (last accessed on 28th June 2015)

Appendix 2.2: Major Research Proposal (V.5)

Title: Body image, self-esteem and other anxieties for Children and Young People with Implantable Cardioverter Defibrillators (ICDs): A Qualitative Study.

Abstract

Background. The development of the Implantable Cardioverter Defibrillator (ICD) has been shown to provide a moderately longer life expectancy for patients who receive an ICD compared to those who do not, reducing the occurrence of heart failure and incidence of sudden cardiac arrest. While the psychosocial effects of an ICD have been well documented with adults, there is limited research on children and adolescents with ICDs.

Aims. This study aims to explore the challenges that young people with ICD devices experience using semi structured, qualitative interviews, with particular reference to its effect on the development of body image and self-esteem in this population.

Methods. Participants will be adolescents aged 11-17 who attend Yorkhill Hospital Paediatric Cardiac Team, Glasgow. Data will be analysed using Interpretive Phenomenological Analysis.

Applications. This study aims to add to the limited evidence base regarding the experiences of adolescents with ICDs, and how this affects their body image, self-esteem and other anxieties. This may help inform the support these patients are offered, in order to optimise the chances of a positive adjustment to having this device.

Introduction

Cardiac arrhythmia, or irregular beating of the heart, can be life threatening in some cases. Fortunately, advances in technology have led to the development of the ICD device. This has been shown to have good outcomes for people who are at risk of life threatening arrhythmias (NICE, 2006). NICOR (2014) reported the rate of ICD implants at 72 per million per year in 2010 in the UK. The vast majority of these are provided to adults between 55-89(NICOR, 2014).An ICD is a small pulse generator which is implanted under the collar bone, which gives small electric pulses to shock the heart into a regular rhythm. The medical benefits of these devices have been documented, providing a moderately longer life expectancy for adults who receive an ICD compared to those who do not, reducing the occurrence of heart failure and incidence of sudden cardiac arrest (Bardy et al., 2005). Three early trials of ICDs with 1850 patients reported a 50% reduction of the risk of cardiac death and a 25-28% reduction of risk of wider cause mortality (Klein et al, 2003; Irvine et al, 2002; Kuck et al, 2000). Bardy et al's (2005) study on 2521 adults with congestive heart failure demonstrated that conventional therapy (CT) plus single shock ICD decreased risk of death by 23% compared to CT plus amiodarone or CT plus a placebo.

More recently there has been a growing interest in the psychological adjustment of patients who have one of these devices fitted. A systematic review of 45 quantitative studies including 5000 adults (Magyar-Russell et al., 2011) suggested a prevalence of 20% for depression and anxiety in adults with an ICD, although rates varied widely from 8-63% for anxiety symptoms and 5-41% for depression. The studies cited used heterogeneous self-report measures, therefore, to obtain more accurate prevalence rates, larger, structured research studies are required. A systematic review

of 22 qualitative studies which interviewed 343 patients and 115 partners and carers, (Palacios-Cena et al., 2011), has identified that adult patients often experience ongoing psychological, social and physical changes and anxieties regarding unexpected electric shocks from the ICD device. Gender differences in adjustment to having an ICD have also been shown, with women identifying more concerns about physical appearance and the impact on social relationships (Palacios-Cena et al., 2011). Exploratory interviews conducted by King et al.(2009), with 13 women with surgery scars identified that the scars were often a powerful reminder of the surgery, which they were not well prepared how to live with following surgery. This study also found that this population reported experiencing lower self-esteem and poor body image. McDonough (2009) explored the experiences of 20 adults aged 18- 40 with ICD devices, who were recruited and interviewed via telephone and the internet. In this population, different ages groups were likely to have different concerns, such as anxiety regarding child bearing and rearing, and financial stability. There is also evidence that adults under 50 years of age consider themselves to be 'different' from peers who do not have health difficulties, and may experience more stress than ICD patients who are 50 years or older (Sears et al., 2001).

Blom (2008) identified that less than 1% of patients with ICDs are under 21 years of age, and there has been little research on how this group of patients adjust to having an ICD. Children and young people with ICD devices will have to negotiate the tasks of growing up and adulthood while coping with their ICD device. Also, this group are likely to have the ICD devices for much longer than older adults who undergo the same surgery, therefore it is important to understand the effects that ICD devices have on this patient group in order to facilitate their adjustment to this life changing

implant. The anxieties and issues that patients with ICD devices experience are likely to vary at different developmental stages. Much is unknown about this groups' experience of having an ICD, and the long term impact it will have on their lives. In a qualitative study of 6 young people aged 12- 17 with ICDs and their parents, Rahman et al. (2012) found common themes for the adolescents with ICDs and their parents, including restrictions imposed on the patients, the shock experience, ongoing challenges post implant and positive benefits of having an ICD. Sears et al (2011) investigated quality of life (QoL) for children and young people with ICDs; they assessed 60 children using self and parent-report measures. This study found that children with ICDs rate their quality of life as comparable to other children with chronic health difficulties, with the exception of lower physical QoL. Girls reported lower QoL than boys, and parents' perception of their children's QoL was lower than the perception of the young people in the study.

Other research has identified that experience of serious health concerns such as cancer can affect the development of positive body image in children and adolescents, and that adolescents are more likely than younger children to have concerns about their appearance (Fan & Eiser, 2009). The effect of having an ICD on body image and self-esteem in children and adolescents has yet to be explored in detail, however may be important in supporting this group to adjust to the device. Concerns regarding body image and self-esteem may be particularly salient where visibility of the device or scar tissue is an issue. Further exploration of the anxieties and challenges of adjusting to life with an ICD may help to inform cardiac teams on how to support this group to adjust successfully post implant.

Aims and Hypotheses

This study aims to explore the anxieties and challenges that adolescents with ICD devices experience, with particular reference to its effect on the development of body image and self-esteem in this population. This may help to inform the development of support that is available to these patients to facilitate optimum adjustment to the device.

Methods

Plan of Investigation

Qualitative interviews will be undertaken with young people who have Implantable Cardioverter Defibrillator devices.

Participants

Participants will be young people aged 11-17 who attend Yorkhill Hospital Paediatric Cardiac Team, Glasgow. Data from the team suggest that there are 13 potential participants who attend the hospital between once a month and once every six months. Potential participants are spread geographically across different health boards in Scotland, from Tayside to Ayrshire and Arran. Eight to ten participants will be recruited for this study and they will be interviewed on a first come first served basis.

Inclusion and Exclusion Criteria

Participants must be fluent English speakers as interviews will be conducted in English. Financial restrictions preclude the use of an interpreter to facilitate interviews in another language. To increase homogeneity of the sample as far as

possible, participants should be aged 11-17. This will attempt to minimise the developmental range of the subject group, while maximising the number of possible respondents out of the limited number available. Unfortunately, as the population of young people with ICDs in Scotland is small, further reduction of the age range may make recruitment unfeasible.

There will be a minimum time requirement of 6 months since implant, in order to allow for adjustment to having an ICD device.

Recruitment Procedures

Possible participants will be contacted by a letter from a member of the Cardiac Team to invite them to take part in the study prior to their hospital appointment. Letters will be used rather than face-to-face contact, to make it easier for potential participants to decline if they so wish. Those who wish to take part will be able to respond using an opt in slip, email or telephone contact. Where hospital appointments are out with the timescale for this study, participants will be invited to attend the hospital for interview, or offered a telephone interview. Parents will be invited to accompany participants, but will be asked to wait in a different room during the interview. All parents will be invited in for a brief discussion with the participant and interviewer after the interview has been conducted, unless the young person does not wish parental feedback to take place. It is anticipated that the study will require a three month period to recruit participants. Unfortunately travel expenses will not be provided to participants due to financial restrictions.

Measures

A semi structured interview will be conducted with each participant. The interview aims to explore participants' perceptions of living with an ICD, as well as the challenges and anxieties they may have experienced, specifically regarding body image and self-esteem. See Appendix III for the draft interview schedule.

Design

This will be a qualitative study comprising of individual interviews, each lasting between thirty minutes to one hour.

Research Procedures

Interviews will ideally be carried out face to face, which is the 'gold standard' in qualitative research (Novik, 2008) at the RHSC, Yorkhill, however telephone interviews may be required to overcome geographical or logistical barriers. The interviews will be digitally recorded and transcribed verbatim, with all patient identifiable data removed. Technical equipment to record telephone interviews is available for this study, and has been used successfully for previous qualitative research at the University of Glasgow. A lay summary of the report will be available on request for parents and participants once the study has been completed, via the cardiac team.

Data Analysis

The data will be explored using Interpretative Phenomenological Analysis (IPA) (Smith, 2009). IPA is an approach which uses semi and unstructured interviews with participants to explore their experiences. IPA is used to understand the significance

of a particular life event for the individual, and the meaning they attach to the event, in the context of their personal and social experiences. IPA also recognises that the researcher's understanding of the participant's experience is affected by the filter of their own experience, knowledge and biases, and this will be acknowledged in the analysis and discussion of the data (Smith, 2009). A cross section of data will be independently analysed by two research supervisors to identify common themes in the data, which will ensure reliability and validity of the findings. Supervision will also be used throughout the analysis to ensure that a plausible and coherent interpretation of the findings is reached, as recommended by Smith et al. (2009).

Data Storage

Recordings will be stored in a password protected file in the NHS GG&C secure drive for the duration of the study and for a limited time after the study is concluded. Transcriptions will be stored on an encrypted University of Glasgow laptop for data analysis, with all identifiable patient information removed. These will be deleted from the laptop once the study is completed, with one copy of the anonymised transcriptions stored in a password protected file on the GG&C secure drive for ten years. Also, one paper copy of each transcription will be stored securely in a locked filing cabinet in the University of Glasgow Academic Unit for Mental Health and Wellbeing. Anonymised transcription data will be transferred from the university laptop to the NHS GG&C secure drive via a secure email account.

Any paper information such as consent forms shall be stored in a locked cabinet in a secure room within the Paediatric Clinical Psychology team base at RHSC, Yorkhill, in accordance with GG&C data protection policy.

Justification of sample size

While the number of participants required for qualitative research vary, guidelines for suggest that between four and ten interviews is an adequate sample size for professional doctorate level research using IPA methodology (Smith et al, 2009). Previous qualitative research in a paediatric clinical psychology population from the same hospital reported high response rates to interviews and identified no barriers to recruitment (Whittaker, 2012), therefore this is not an anticipated difficulty for this study.

Settings and Equipment

Participants will be interviewed at the Royal Hospital for Sick Children, Yorkhill, Glasgow where possible, or via telephone at the same site. A digital voice recorder will be required, as will amenities for participants. A laptop and headphones will be required to transcribe the interviews. Funding for all research costs has been approved and will be provided by the University of Glasgow.

Health and Safety Issues:

These have been discussed in more detail in Appendix II.

Researcher Safety Issues

Interviews will be conducted on site at Yorkhill, or by telephone interview. Medical care will be accessible on site. There will be no lone working, and the researcher will be subject to regular supervision during the project.

Participant Safety Issues

The participants will be able to stop the interview at any time, and breaks will be offered throughout. In the event of distress, the participant will be supported by the researcher (Trainee Clinical Psychologist), who receives supervision from a qualified clinical psychologist. Interviews will be conducted during normal working hours.

Ethical Issues

In accordance with the ScotCRN guidance on obtaining consent for children and young people in Scotland, participants under the age of 16 will be asked to give informed consent if they are deemed to have capacity to do so by a qualified healthcare professional. Assent from a parent/ guardian will also be sought for this participant group. Participants aged 16 and over are able to provide informed consent themselves. Research procedures for working with children will be in line with guidance from the 2010 Code of Human Research Ethics (BPS, 2010) and National Children's Bureau (2003). The limits of confidentiality will be discussed at the beginning of the interview, and confidentiality procedures will be followed should information concerning child protection be raised during the interview process. Emotional support will be provided if there is distress, and the interview will be terminated if required. Participants will be made aware they can stop the interview or withdraw consent at any time and this will not affect their relationships with the Cardiac Team or their future care. Follow-up support will be offered through the Department of Paediatric Clinical Psychology at the Royal Hospital for Sick Children with the specialist psychologist attached to the Cardiac Team. All data will be anonymised on transcription. NHS GG&C Data protection policy and BPS

guidelines on data protection for Clinical Psychologists will be followed throughout the study. Recordings will be destroyed after transcriptions have been completed.

Financial Issues

This study will require finance to cover equipment and administration costs. Most equipment will be available for use through the University of Glasgow and administration costs should be limited to printing and postage. See Appendix I for more a more detailed breakdown of costs.

Timetable

See Appendix VII for more detail.

Timetable for MRP	
Month	Task
December 2013	Outline for MRP
January 2014	Draft proposal for MRP handed in
April 2014	Final proposal for MRP handed in
May 2014	Final proposal marked and changes identified (3 weeks). Changes made and re-submitted.
May 2014-	Apply for R & D ethics (3-4 weeks). Consent form and participant information sheets completed for this.
June	R&D approval. REC application submitted. 4-8 weeks for

	REC approval, possible extra time required for changes -2 weeks.
August/ September 2014	REC approval
October 2014	Begin recruitment process
November 2014- March 2015	Data collection- 8 hours interview. Collect within 3 months (Nov-Jan) and begin transcription as it proceeds. Transcription- roughly 40 hours. IPA analysis Jan 2015-Mar 2015.
May 2015	First draft of MRP submitted
July 2015	Final MRP submitted

Practical Applications

This study aims to add to the limited evidence base regarding the experiences of children and adolescents with ICDs, how this affects their body image, self-esteem and other anxieties. This will help to inform cardiac teams what issues this patient group faces, which is likely to be different for the issues faced by adults with this implant. This may help inform the emotional and psychological support these patients are offered, in order to optimise the chances of a positive adjustment to having this device. For example, results may help shape preparatory work offered to young people and their families before the young person undergoes an ICD procedure, as well as the after-care support offered by the team. As Clinical Psychology support to cardiology at the RHSC is a relatively new service, user feedback would be welcome to help prioritise the direction of care and inform patient need.

References

- Bardy, G., Lee, K., Mark, D., Poole, J, Packer, D.,Boineau, R., Domanski, M., Troutman, C., Anderson, J., Johnson, G., McNulty, S., Clapp-Channing, N., Davidson-Ray, L., Fraulo, E., Fishbein, D.,Luceri,R., Ip, J., (2005) Amiodarone or an implantable cardioverter-defibrillator for congestive heart failure. *The New England Journal of Medicine*. 352 (3). p225-237
- Blom NA. (2008) Implantable cardioverter-defibrillators in children. *Pacing and Clinical Electrophysiology*. 31. p32–34.
- The British Psychological Society (2010).*Code of Human Research Ethics*. The British Psychological Society: Leicester
- DiCicco-Bloom, B.,& Crabtree, B. (2006) The qualitative research interview. *Medical Education*,40 (4). p 314–321
- Fan, S., &Eiser, C., (2009) Body image of children and adolescents with cancer: A systematic review. *Body Image*. 6 , p247-256.
- Irvine, J., Dorian, P., Baker, B., O'Brien, B., Roberts, R., Gent, M., Newman, D., & Connolly, S., (2002) Quality of life in the Canadian implantable defibrillator study (CIDS). *American Heart Journal*.144 (2). p282-289.
- Klein, R., Raitt, M., Wilkoff, B., Beckman, K., Coromilas, J., Wyse, G., Friedman, P., Martins, J., Epstein, A., Hallstrom, A., Ledingham, R., Belco, K., &Greene, L.,(2003). Analysis of implantable cardioverter defibrillator therapy in the antiarrhythmics versus implantable defibrillators (AVID) trial. *Journal of Cardiovascular Electrophysiology*.14 (9). p940-948.
- King, K., McFetridge-Durdle, J., LeBlanc, P., Anzarut, A &Tsuyuki, R. (2009). A descriptive examination of the impact of sternal scar formation in women. *European Journal of Cardiovascular Nursing*. 8. p112-118.

Kuck,K., Cappato, R., Siebels, J., Ruppel,R., (2000). Randomized comparison of antiarrhythmic drug therapy with implantable defibrillators in patients resuscitated from cardiac arrest: The cardiac arrest study Hamburg (CASH). *Circulation*.102. p748-754

Magyar-Russell, G., Thombs, B., Cai, J., Baveja, T.,Kuhl, E., Singh, P., Barroso, M., Arthurs, E., Roseman, M., Amin, N., Marine,J. &Ziegelstein, R. (2011). The prevalence of anxiety and depression in adults with implantable cardioverter defibrillators: A systematic review. *Journal of Psychosomatic Research*. 71. p223-231.

McDonough, A (2009) The experiences and concerns of young adults (18-40 years) Living with an implanted cardioverter defibrillator (ICD). *European Journal of Cardiovascular Nursing*.8. p274- 280.

National Children's Bureau (2003). *Guidelines for research*. London: NCB.

National Institute for Cardiovascular Outcomes Research (NICOR) <http://nicor4.nicor.org.uk/> [accessed on 21st February 2014.]

NHS Greater Glasgow And Clyde. *NHS Data Protection policy*. Glasgow: NHS GG&C

NHS Greater Glasgow And Clyde . *Confidentiality policy*. Glasgow: NHS GG&C.

National Institute for Health and Clinical Excellence (Jan 2006) Implantable cardioverter defibrillators for arrhythmias. *Review of Technology Appraisal 11*. NICE: London www.nice.org.uk

Novick, G., (2008). Is there a bias against telephone interviews in qualitative research? *Research in Nursing & Health*. 31. p391–398.

Palacios-Cena, D., Losa-Iglesias, M., Alvarez-Lopez, C., Cachon-Perez, M., Reyes, R., Salvadores-Fuentes, P. & Fernandes-de-las-Penas, C. (2011). Patients, intimate

partners and family experiences of implantable cardioverter defibrillators: qualitative systematic review. *Journal of Advanced Nursing*. 67 (12). p 2537-2550.

Rahman, B., Macciocca, M., Sahhar, M., Kameri, S., Connell, V., & Duncan, R. (2012) Adolescents with implantable cardioverter defibrillators: A patient and parent perspective. *Pacing and Clinical Electrophysiology*. 35 (1). p62- 72 .

Sears, SF., Burns, J.L., Handberg, E., Sotile, W.M., Conti, J.B.(2001) Young at heart: understanding the unique psychosocial adjustment of younger implantable cardioverter defibrillator recipients. *Pacing and Clinical Electrophysiology*.24. p1113-1117.

Sears, S., Hazelton, G., St. Amant, J., Matchett, M., Kovacs, A.,m Vazquez, L., Fairbrother, D., Redfearn, S., Hanisch, D., Dubin, A., Cannon, B., Fishbach, P.,Kanter, R., & Bryant, R (2011). Quality of life in paediatric patients with implantable cardioverter defibrillators. *American Journal of Cardiology*.107. p1023–1027.

Smith, J., Flowers, P.,& Larkin, M., (2009) *Interpretative Phenomenological Analysis: Theory, Method and Research*. SAGE Publications Ltd: London.

Whittaker, J.A (2012) The effect of insulin pump therapy on children and adolescents' quality of life: A qualitative study. Doctorate of Clinical Psychology thesis. University of Glasgow thesis service (unpublished).

Appendix 2.3: Health and Safety Form (MRP V.5)

WEST OF SCOTLAND/ UNIVERSITY OF GLASGOW
DOCTORATE IN CLINICAL PSYCHOLOGY
HEALTH AND SAFETY FOR RESEARCHERS

1. Title of Project	Body image, self esteem and other anxieties for Children and Young People with Implantable Cardioverter Defibrillators (ICDs): A Qualitative Study.
2. Trainee	
3. University Supervisor	Dr Alison Jackson
4. Other Supervisor(s)	Dr Sarah Wilson
5. Local Lead Clinician	Dr Kathleen McHugh
6. Participants: (age, group or sub-group, pre- or post-treatment, etc)	Young people aged between 11 and 17 years old who attend Yorkhill Hospital Paediatric Cardiac Team who have an Implantable Cardioverter Defibrillator (ICD). Between 8-13 potential participants. Target sample: 8 participants.
7. Procedures to be applied (eg, questionnaire, interview, etc)	Semi structured, individual interviews lasting between 30mins -1 hour depending on the participant.
8. Setting (where will procedures be carried out?) i) General	Participants will be interviewed at Yorkhill Paediatric Hospital where possible, or via telephone at the same site.
ii) Are home visits involved	No
9. Potential Risk Factors Identified (see chart)	<p>The participants are children and adolescents with chronic health difficulties who are familiar with regular contact with health services. They have established relationships with the cardiac team and are well known by the service. They are not associated with dangerous or unpredictable behaviour.</p> <p>Participants may become tired or unwell as a result of their health difficulties.</p> <p>Participants may become distressed during the</p>

	<p>interview, although the procedures used in the study are similar to those used by clinical psychologists in previous research.</p> <p>The interview schedule is not designed to cause any alarm or distress.</p> <p>Researcher and participant safety in the environment: The interviews will be conducted in a clinical research setting that participants routinely attend. There are specific procedures in place at the Royal Hospital for Sick Children to minimise risk to staff and these are thought to be adequate in the context of the proposed study, such as having colleagues in the vicinity who can respond with support if required.</p> <p>Confidentiality: If information is disclosed which raises concerns about the participants welfare, or the safety of another individual, confidentiality may be broken and standard NHS GG &C procedures regarding child protection will be followed.</p>
<p>10. Actions to minimise risk (refer to 9)</p>	<p>There will be access to medical attention if required during the interview process, should health concerns arise. Comfort breaks will be provided where needed, and the participant or their parent will be advised that they are able to stop the interview at any time.</p> <p>Participants will be advised to bring a responsible adult with them for support (although they may be asked to wait outside if the participant is comfortable with this). This will allow the participant privacy to discuss worries openly without the potential influence of a parental presence.</p> <p>While the interviews will not be designed to cause any distress to the participant, should this occur during the course of the interview the participant will be supported by the researcher (Trainee Clinical Psychologist), who receives supervision from a qualified clinical psychologist.</p> <p>Participant and researcher safety in the environment: There will always be other staff in the building, and the researcher will be able to</p>

	<p>contact supervisors if required. Regular supervision by a qualified clinical psychologist will support researcher with any distressing information which they may be exposed to.</p> <p>Interviews will be conducted during normal working hours.</p> <p>Confidentiality: Informed consent will be sought from participants and their parents for the interviews. The limits of confidentiality will be discussed at the beginning of the interview, and confidentiality procedures will be followed should concerning information regarding the child be raised during the interviews. Adequate emotional support to be provided if there is any distress, and the interviews will be terminated at this point</p>
--	--

Trainee signature:Date:

University supervisor signature:..... Date:

Appendix 2.4: Research Costs

RESEARCH EQUIPMENT, CONSUMABLES AND EXPENSES

Trainee

Year of Course ...Second year.....

Intake Year.....2012.....

Please complete the list below to the best of your ability:

Item	Details and Amount Required	Cost or Specify if to Request to Borrow from Department
Stationary	Pens and highlighters.	Owned by researcher
Postage	Postage for recruitment letters and summary of results at end of study. 2 nd class Stamps x 40	40x 50p= £20.00
Photocopying and Laser Printing (includes cost of white paper)	Printing costs for letters, information sheets, transcripts of interviews	1x Ream of paper= £2.80
Equipment and Software	Digital voice recorder, Headphones, telephone recorder, laptop, NVivo software.	Borrow from department £30 for department license
Measures	Semi structured interview- no copyright measures required.	0.00
Miscellaneous	Refreshments- tea, coffee, milk, juice, biscuits, sweets. Tissues. Phone bill for telephone interviews	£30.00 NHS GG& C

Trainee Signature..... Date.....

Supervisor's Signature Date

Appendix 2.5: Plain English Summary (MRP V.5)

Title: Body image, self-esteem and other anxieties for Children and Young People with Implantable Cardioverter Defibrillators (ICDs): A Qualitative Study.

Background

The development of the Implantable Cardioverter Defibrillator (ICD) has been shown to have good outcomes for people who are at risk of life-threatening irregular heart rhythms. Studies suggest that ICDs give a longer life expectancy for patients, reducing the occurrence of heart failure and sudden cardiac arrest. An ICD is a small pulse generator which is implanted under the collar bone, which gives small electric pulses to shock the heart into a regular rhythm. Research about the emotional impact of having an ICD suggests that 20% of adult patients suffer from depression and anxiety (Magyar-Russell et al., 2011). Other research suggests that patients often experience ongoing psychological, social and physical changes after getting an ICD. Very little research has been done looking at the experience of children and adolescents with ICDs (Rahman et al., 2012). The effect of having an ICD on body image and self-esteem in children and adolescents is unknown, however may be important in supporting this group to adjust to the device.

Aims

This study aims to explore the experiences of young people with ICD devices, particularly its effect on their body image and self-esteem. This may help to develop the support that is available to these patients to help them to adjust to having an ICD as successfully as possible.

Methods

Semi structured interviews will be undertaken with children and adolescents who have Implantable Cardioverter Defibrillator devices. Interviews will last between 30mins -1 hour.

Participants

Participants will be young people aged 11-17 who attend Yorkhill Hospital Paediatric Cardiac Team.

Recruitment

Participants will be contacted by letter to explain the study before their hospital appointment by a member of the cardiac team. They will then be contacted again for consent to interview by follow up letter.

Consent

Informed consent will be given by both participants and their parents for the interviews.

Study design

A semi structured interview will be carried out with each participant.

Data collection

The interviews will be recorded and typed up (transcribed) word for word, with all patient identifiable data removed. Recordings will then be destroyed. The transcribed information will then be stored according to NHS GG&C Data Protection Policy.

Ethical Issues

Confidentiality will be discussed in interview, and procedures will be followed if there are concerns about the child's welfare during the interview. Emotional support will be provided immediately if needed and the interviews will be stopped. Follow up support will be offered through the Paediatric Cardiac team at Yorkhill. It will be explained that they can stop the interview or withdraw consent at any time and this will not affect their relationships with the cardiac team.

Practical Applications

This study aims to add to the limited evidence base regarding the experiences of children and adolescents with ICDs. This may help inform the support these patients are offered, in order to support their adjustment to having this device.

References

Magyar-Russell, G., Thombs, B., Cai, J., Baveja, T., Kuhl, E., Singh, P., Barroso, M., arthurs, E., Roseman, M., Amin, N., Marine, J. & Ziegelstein, R. (2011). The prevalence of anxiety and depression in adults with implantable cardioverter defibrillators: A systematic review. *Journal of Psychosomatic Research*. 71, 223-231.

Rahman, B., Macciocca, M., Sahhar, M., Kameri, S., Connell, V., & Duncan, R. (2012) Adolescents with Implantable Cardioverter Defibrillators: A Patient and Parent Perspective. *Pacing and Clinical Electrophysiology*, 35 (1), 62- 72

Word Count: 576

Appendix 2.6: Research Ethical Approval Letters

WoSRES
West of Scotland Research Ethics Service



West of Scotland REC 3

Ground Floor – The Tennent Institute
Western Infirmary
38 Church Street
Glasgow G11 6NT
www.nhs.gov.uk

Dr Alison Jackson
University Teacher
University of Glasgow
Mental Health and Wellbeing, Institute of
Health and Wellbeing
Admin building, Gartnavel Royal Hospital
1055 Great Western Road
Glasgow G12 0XH

Date 15th October 2014
Your Ref
Our Ref
Direct line 0141 211 2123
Fax 0141 211 1847
E-mail WOSREC3@ggc.scot.nhs.uk

Dear Dr Jackson

Study title:	Body image, self-esteem and other anxieties for Children and Young People with Implantable Cardioverter Defibrillators (ICDs): A Qualitative Study.
REC reference:	14/WS/1082
IRAS project ID:	154720

Thank you for responding to the Committee's request for further information on the above research and submitting revised documentation. The further information has been considered on behalf of the Committee by the Chair and Alternate Vice Chair.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to make a request to postpone publication, please contact the REC Manager, Mrs Liz Jamieson, wosrec3@ggc.scot.nhs.uk.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at <http://www.rdforum.nhs.uk>.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of approvals from host organisations

Ethical review of research sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Interview schedules or topic guides for participants [Interview Schedule]	6	25 July 2014
IRAS Checklist XML [Checklist_03102014]		03 October 2014
IRAS Checklist XML [Checklist_15082014]		15 August 2014
Letter from funder [University approval]		30 June 2014
Letters of invitation to participant [Invitation to project-parent]	7	18 September 2014
Letters of invitation to participant [Invitation to project-child]	7	18 September 2014
Other [Research nurse CV]		11 August 2014
Other		11 August 2014
Other [Clinical research supervisor CV]		11 August 2014
Other [Equipment and funding]		11 August 2014
Other [Covering letter for Ethics Changes]	1	18 September 2014
Other [Health and Safety form]		11 August 2014
Participant consent form [Parent Assent Form]	6	25 July 2014
Participant consent form [Participant Consent Form]	6	25 July 2014
Participant consent form [Child Assent form]	6	25 July 2014
Participant consent form	6	25 July 2014
Participant information sheet (PIS)	7	18 September 2014
Participant information sheet (PIS) [PIS 16-17]	7	18 September 2014
Participant information sheet (PIS) [PIS 11-15]	7	18 September 2014
Participant information sheet (PIS) [Parent Information Sheet]	6	25 July 2014

Participant information sheet (PIS) [Participant Information sheet]	6	25 July 2014
REC Application Form [REC_Form_15082014]		15 August 2014
Research protocol or project proposal [MRP Proposal]	6	25 July 2014
Summary CV for Chief Investigator (CI) [AJ CV]		06 August 2014
Summary CV for student [LC CV]		07 August 2014
Summary CV for supervisor (student research) [SW CV]		06 August 2014
Summary, synopsis or diagram (flowchart) of protocol in non technical language [Plain English Summary]	6	25 July 2014

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document "*After ethical review – guidance for researchers*" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website:

<http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/>

HRA Training

We are pleased to welcome researchers and R&D staff at our training days – see details at

<http://www.hra.nhs.uk/hra-training/>

14/WS/1082	Please quote this number on all correspondence
-------------------	---

With the Committee's best wishes for the success of this project.

Yours sincerely



Liz Jamieson
REC Manager
On behalf of Dr Adam Burnel, Chair

Email: wosrec3@ggc.scot.nhs.uk

Enclosures: "After ethical review – guidance for researchers"

Copy to: *Ms Emma-Jane Gault, University of Glasgow*
Miss Joanne McGarry, Academic Research Co-ordinator, Research and Development, NHS GGC

Coordinator: Mrs Joanne McGarry
Telephone Number: 0141 211 2142
E-Mail: joanne.mcgarry@ggc.scot.nhs.uk
Website: www.nhsggc.org.uk/r&d

R&D Management Office
Western Infirmary
Tennent Institute
1st Floor 38 Church Street
Glasgow, G11 6NT,

21 October 2014

Dr Alison Jackson
University Teacher
Institute of Health and Wellbeing
Admin Building
Gartnavel Royal Hospital
1055 Great Western Road
Glasgow G12 0XH

NHS GG&C Board Approval

Dear Dr Jackson,

Study Title:	Body image, self-esteem and other anxieties for Children and Young People with Implantable Cardioverter Defibrillators (ICDs): A Qualitative Study.
Principal Investigator:	Dr Alison Jackson
GG&C HB site	Royal Hospital for Sick Children
Sponsor	NHS Greater Glasgow and Clyde
R&D reference:	GN14KH437
REC reference:	14/WS/1082
Protocol no:	V6; 25/07/14

I am pleased to confirm that Greater Glasgow & Clyde Health Board is now able to grant **Approval** for the above study.

Conditions of Approval

1. **For Clinical Trials** as defined by the Medicines for Human Use Clinical Trial Regulations, 2004
 - a. During the life span of the study GGHB requires the following information relating to this site
 - i. Notification of any potential serious breaches.
 - ii. Notification of any regulatory inspections.

It is your responsibility to ensure that all staff involved in the study at this site have the appropriate GCP training according to the GGHB GCP policy (www.nhsggc.org.uk/content/default.asp?page=s1411), evidence of such training to be filed in the site file.

2. **For all studies** the following information is required during their lifespan.
 - a. Recruitment Numbers on a monthly basis
 - b. Any change of staff named on the original SSI form
 - c. Any amendments – Substantial or Non Substantial
 - d. Notification of Trial/study end including final recruitment figures
 - e. Final Report & Copies of Publications/Abstracts

Please add this approval to your study file as this letter may be subject to audit and monitoring.

Your personal information will be held on a secure national web-based NHS database.

I wish you every success with this research study

Yours sincerely,

A black rectangular redaction box covering the signature of Mrs Joanne McGarry.

Mrs Joanne McGarry
Research Co-ordinator

Cc: Student – Mrs Louisa Casselden

Appendix 2.7: Information Pack



University
of Glasgow



University of Glasgow
Mental Health and Wellbeing
Gartnavel Royal Hospital
Administration Building
1055 Great Western Road
Glasgow, G12 0XH

Participant information Sheet: 11-15 years old

Title: Body image, self-esteem and other anxieties for Children with Implantable Cardioverter Defibrillators (ICDs).

We would like to ask you to take part a research study. Before you decide if you want to take part, you need to know why the study is being done, and what you would need to do. Please read this sheet carefully and ask any questions you might have and, if you want, talk about it with other people. We will do our best to explain it and you can ask more questions at any time.

Who is carrying out the research?

The research is being carried out by Louisa Casselden, Trainee Clinical Psychologist, with Dr Alison Jackson and Dr Sarah Wilson from Glasgow University. Dr Kathleen McHugh from the Clinical Psychology Department at the Royal Hospital for Sick Children, will also be helping with the project.

Why are we doing this study?

We want to find out about what it is like for children and young people living with ICDs, and if they have any worries about it. This information may help the hospital to look after children with ICDs, to help them to live a happy and full life. The project is being carried out as part of the Doctorate in Clinical Psychology qualification at the University of Glasgow.

Why have I been invited to take part?

Young people who are 11-17 years old and have an ICD and have been asked if they would like to take part in the study. About 8-10 young people will be interviewed. Once we have this many we will not interview anyone else.

Do I have to take part?

No- it is up to you and your parent/ guardian to decide if you want to take part. If you decide you do want to take part you will get a copy of this sheet to keep and you will be asked to sign a form to show you are happy to take part. Your parent/

guardian will also sign a form to say this is OK. You can change your mind at any point and you do not have to give us a reason for this. It will not affect your treatment at all.

What will happen if I take part?

You will meet with Louisa Casselden to talk about what it is like living with an ICD and how it affects your life. This interview will take about 30-60 minutes and will be recorded. The interview will take place at the Royal Hospital for Sick Children, Yorkhill. If you cannot come to the building for an interview we can speak over the telephone. It is a good idea to bring a parent or responsible adult with you, and we will ask them to wait in another room while we talk. You can take a break or stop the interview at any time.

The interview will be kept private; we will not tell your doctor or parents about what we talk about, unless we are worried about your safety. Your information and anything you say may be looked at by the organisers of the study to make sure it is being done properly. The recording will be destroyed after everything that was said has been written down. Your name and other personal information will be taken out. The information collected in the study will be written up in a report, and handed in to the University of Glasgow. Direct quotations may be used in the final report and presentations or publications. People who take part can get copies of the findings of the study once it is finished, either by post or email depending on what you prefer.

Are there any possible risks involved with taking part?

There are no risks involved with taking part, although some people may not want to talk about how they feel. If we were worried we would ask if you would like extra help from the Cardiac team or from the psychologist attached to the Cardiac team. We would talk about this with your parent/ guardian.

Who has reviewed this study?

This study has been reviewed and approved by the University of Glasgow, the NHS West of Scotland Research Ethics Committee and the NHS Greater Glasgow and Clyde Research and Development Department.

If you are interested in taking part?

If you and your parent/ guardian are happy for you to take part, please fill in the tear off slip on the information sheet and send it back to us either in the envelope provided (*no stamp required*), or give it to a member of the RHSC Paediatric Cardiac Team.

Or please contact Louisa Casselden at louisa.casselden@ggc.scot.nhs.net or Dr Alison Jackson on 0141 211 3920.

If you have any more questions about this study, please feel free to contact us. Or, if you would like to talk to an someone who is not in research team, please contact Dr Hamish McLeod at the University of Glasgow, on 0141 211 3920.

Thank you for reading this. Any information you can give us about your experience would be very helpful for us, so that we can make sure we are providing the best service we can to children and young people with ICDs.

Thank you for taking the time to read this Information Sheet and for thinking about taking part in this study

Tear off slip)-----

Please return to: Mrs Louisa Casselden/ Dr Alison Jackson, University of Glasgow, Mental Health and Wellbeing Gartnavel Royal Hospital, Administration Building, 1055 Great Western Road, Glasgow, G12 0XH

Research Study: Body image, self-esteem and other anxieties for Children and Young People with Implantable Cardioverter Defibrillators (ICDs)

Chief Investigator: Dr Alison Jackson, Academic Tutor (University of Glasgow/ NHS Greater Glasgow and Clyde)

Name of young person:

Name of Parent/Guardian:

Signature:

Telephone:

For office use: Body image, self-esteem and other anxieties for Children and Young People with Implantable Cardioverter Defibrillators (ICDs): A Qualitative Study

Participant number:

Cardiology Team
Royal Hospital for Sick
Children
Yorkhill,
Glasgow,
G3 8SJ
25th July 2014

Dear (*name*),

I would like to invite you take part in a study which I am carrying out with the Paediatric Cardiac team at the Royal Hospital for Sick Children (RHSC), Yorkhill Hospital, as part of the Doctorate in Clinical Psychology qualification from the University of Glasgow. The purpose of this study is to find out about the experiences of young people who have an Implantable Cardioverter Defibrillator.

A leaflet is enclosed with more information about the study. Before you make up your mind, it is important for you to understand what we are asking you to do and why. Please take time to read the enclosed information sheet carefully, and feel free to talk about it with other people. If you would like any more information about it you can contact me directly, or Sharon Watson, Cardiac Liaison Nurse on 0141 201 9291.

If you would like to take part in the study please return the tear off slip in the envelope provided. Alternatively you can let us know using our email or telephone details. You can say no if you don't want to take part, and you can change your mind at any time. This will not affect the clinical care you get from the team in any way.

Yours sincerely,

Louisa Casselden

Dr Karen McLeod

Dr Kathleen McHugh

Trainee Clinical
Psychologist

Consultant Cardiologist

Principal Clinical Psychologist

University of Glasgow

RHSC, Yorkhill

RHSC, Yorkhill

Appendix 2.8: Consent and Assent Forms



University of Glasgow
Mental Health and Wellbeing
Gartnavel Royal Hospital
Administration Building
1055 Great Western Road
Glasgow, G12 0XH

Subject Number:

Body image, self-esteem and other anxieties for Children and Young People with Implantable Cardioverter Defibrillators (ICDs): A Qualitative Study

Participant **Consent Form**

Please

Initial box

1.	I have read and understood the information sheet dated 25/07/2014 (version 6). I have had time to think about the project and I have been able to ask questions and have them answered.	
2.	I understand that my participation is voluntary and I can stop the interview at any time, and this will not affect the medical care I receive.	
3.	I know that the interview will be recorded by the researcher, Louisa Casselden and only be used for the purposes of the research study, as described in the information sheet. I understand that my information may be looked at by the study organisers to make sure it's being run properly.	
4.	I agree that comments from my interview can be used in the final report, presentations and publications. All names, places and any patient identifiable information will be removed. Nothing that identifies me will appear for others to see.	
5.	I agree to take part in the above study.	

6.	I would like a summary of the research project to be sent to me at the end of the project by the Cardiac team. I can get more copies of the summary by letting the team know I would like them.	
----	---	--

Name of child/young person:

Name of person giving consent: _____

Signature: _____ Date: _____

Contact Tel No: _____

Name of person taking consent:

Signature: _____ Date: _____

When complete: 1 for participant; 1 for researcher site file; 1 (original) to be kept in medical notes.



University
of Glasgow



University of Glasgow
Mental Health and Wellbeing
Gartnavel Royal Hospital
Administration Building
1055 Great Western Road
Glasgow, G12 0XH

Subject Number:

Project Title: Body image, self-esteem and other anxieties for Children and Young People with Implantable Cardioverter Defibrillators (ICDs): A Qualitative Study

ASSENT FORM FOR PARENTS

(To be completed by parent/guardian)

Have you read the information about this project?	Yes/No
Has somebody else explained this project to you?	Yes/No
Do you understand what this project is about?	Yes/No
Have you asked all the questions you would like to?	Yes/No
Have you had your questions answered?	Yes/No
Do you understand that your child can stop taking part at any time?	Yes/No
Are you happy for your child to take part?	Yes/No

If any answers are 'no' or you don't want your child to take part, don't sign your name!

If you are happy for your child to take part please sign your name below:

Your name _____

Signature _____

Date _____

Name of person taking assent:

Print Name _____

Signature _____

Date _____

Thank you for your help

When complete: 1 for participant; 1 for researcher site file; 1 (original) to be kept in medical notes.

Appendix 2.9: Interview Schedule

This interview schedule was designed by the interviewer and was informed by recommendations from the literature (DiCicco-Bloom & Crabtree, 2006; Smith et al., 2009), a previous doctoral thesis in paediatric clinical health (Whittaker, J., 2012), and through discussion with academic supervisors at The University of Glasgow.

Interview Schedule

The below questions will be used as a guide to instigate discussion. At the beginning of the interview participants will be reminded of the boundaries of confidentiality, and will be invited to use the amenities provided as required. They will be reminded that the interviews will be anonymised, and they can take a break or stop the interview at any point. Also, it will be made clear that this is a discussion, and there are no right or wrong answers.

A. Demographic Information

Age; Gender; Number of months since ICD device was implanted; How often patient attends cardiac clinic

B. Living with an ICD device

Q: What was it like for you when you found out you needed an ICD?

Prompt: What do you think about having an ICD?

Q: What is it like living with an ICD?

Prompt: How does it affect your day to day life?

Q: What does the ICD do for you?

Prompt: Has this ever been explained to you? How did they explain it/ How would you describe it?

C. Changes in having an ICD

Q: Is there anything that you have noticed has changed since you got an ICD?

Prompt: what was life like before the ICD? Is that still the same now? Can you do the same things you did before having the ICD? Do people treat you the same?

Q: Is your life better or worse than your friends? (Q of L in having ICD?)

Prompt: Can you do the same things as your friends? Play games/ take part in activities? Activities/ school and home routines?

Q: Does having the ICD stop you doing anything or help you to do anything you couldn't do before?

Q: Are there changes in the way you feel physically since getting the ICD?

Q: Are there any changes in your feelings since getting an ICD?

Prompt: E.g. more happy/sad/worried/calm

Q: Has having an ICD affected you socially?

D. Body image, self-esteem and anxiety regarding ICD

Q: Do you have any worries about having an ICD?

Prompt: What is it that worries you?

Q: Do other people know you have an ICD? Would you tell them?

Prompt: why do you tell/not tell them? Do you worry about that?

Q: Has it affected the way you think about yourself?

Prompt: confidence/ identity/ how you think about your body?

Q: Do you think the ICD has affected the way you look?

Prompt: Clothes you wear?

Q: Do you have a scar from the ICD? What do you think of it?

Q: Has the way you feel about yourself changed since getting an ICD?

Q: Have you experienced a shock from your ICD?

Prompt: If yes, what was that like? Do you worry about that happening again?

E. What has helped with having an ICD?

Q: What support did you get from the hospital and other people?

Prompt: e.g. someone to talk to, opportunities to ask questions, directed to info on the internet, information leaflets.

Q: What has helped you get used to living with having the ICD?

Prompt: attitudes of parents/carers/ hospital staff?

F. Thoughts on the Future

Q: How do you see yourself in the future?

Prompt: what would you like to be in the future?

Q: Do you think having an ICD might affect what you do in the future?

G. Benefits and costs of having an ICD

Q: Do you think there are the good things about having an ICD?

Q: Are there any down sides to having an ICD?
e.g. Reactions from parents and peer group.

H. Overall

Q: Is there anything else you would like to tell me?

Appendix 2.10: Excerpt from transcript analysis

<p>Uncertainty about the future</p> <p>Treatment burden</p> <p>Carrying on with life</p> <p>Development over time</p> <p>Dilemma, conflicting feelings/thoughts</p>	<p>P: And I think it just got, like obviously <u>I'm fine right now</u>, but <u>at the time</u> when it was like, coming in all the time, it was constant like, em, like, right, we are gonna need to keep you in for this, so I think that got to me as well, it was like, I just want to get on with my life, but <u>at the same time</u>, as I say, its like, for my own good.</p>	<p><i>Difficulty finding the words.</i> Health has improved, but uncertainty of future-focus on the moment. Change from before when he was physically unwell. Passage of time, development . Constant difficulty, endurance. Conflicting thoughts, simultaneous. Inner conflict- life giving vs impeding life. Affected his emotions <i>Sentence very long, reflecting the constant stream of events?</i> Determination to get on with life</p>	<p>108</p> <p>109</p> <p>110</p> <p>111</p>
	<p><i>I: Ok, yeah. So, so, you said you think that when you went back to, what was it like going back to school and things after you got it?</i></p>		<p>112</p> <p>113</p>
<p>Journey of ICD</p> <p>Settling in</p> <p>Limitations</p>	<p>P: it was weird [laughs]em, it was, it was as if like, you were <u>coming back, like, coming back</u> and settle in I think again after I had been off, so as well, so, em, and obviously so teachers had to find out about <u>what I had, what I had and like, all the stuff I could, can and can't do. As well, em.</u></p>	<p><i>Use of humour.</i> Unusual experience. Like being on a journey and returning home? Back to normal routine, old life. Settling in period. Describes telling people about iCD. Sharing knowledge. Limitations . Feels like an additional difficulty to cope with after traumatic events <i>Discusses limitations in past and present tense- ongoing.</i></p>	<p>114</p> <p>115</p> <p>116</p> <p>117</p>
	<p><i>I: And did you, were you able to give them that information or did they get that from</i></p>		<p>118</p>

	<i>the hospital or from your parents?</i>		
Physical ill health	P: I gave them some but because <u>I was still quite ill</u> , but I think most of it came from either my mum or from the hospital, ‘n obviously my GP probably. Hmm	Still physically unwell. Sources of information	119 120
	<i>I: Ok, so what’s it like now, living with having the ICD device?</i>		121
Development over time. Constant impact. Limitations Dilemma/conflicting desires Knowledge as coping strategy-rationalising. Coping strategy-positive reframing, minimising Carrying on with life Supported by ICD	P: Now? <u>Now obviously I am more aware of what’s going on and stuff</u> , and <u>I know what I can and can’t do</u> , as I say, <u>I can’t</u> , I shouldn’t be going on, like, rides and I <u>can’t</u> do what, <u>I can’t</u> do contact sport, or <u>can’t</u> jump into a swimming pool, em, <u>I can’t</u> be next to , like, loudspeakers and stuff, em, and as well, <u>I can’t</u> em, and stuff like that, that’s , <u>so I know all the</u> do’s and don’ts. Em, but, <u>as I say, there’s, there’s still always this thing</u> that ‘oh I really want to do that’ but at the same time, <u>its not as if I am missing that much, it’s not that much that’s missed out</u> , now I <u>just get on wi’ it</u> , there’s always that worry that, as I say, it’s like I know <u>it’s in good hands</u> , no matter what happens.	Development of knowledge over time. Limitations- <i>repetition of ‘can’t’ emphasising the number of restrictions.</i> <i>Cumulative effect.</i> Knowledge Conflicting- desire to do things that he can’t Constant, ongoing impact of limitations rationalising, talking himself round, being positive- minimising the impact of limitations. <i>Positive reframing.</i> Getting on with life Feeling supported, responsibility held by someone/something else? ICD?	122 123 124 125 126 127 128 129 130

