Burns, Jennifer (2023) *Understanding the impact of chronic health conditions in children and young people, using qualitative methods.* D Clin Psy thesis.

https://theses.gla.ac.uk/83745/

Understanding the impact of chronic health conditions in children and young people, using qualitative methods

Jennifer Burns

B.A. (Hons), MSc

Submitted in partial fulfilment of the requirements for the degree of

Doctorate in Clinical Psychology

School of Health and Wellbeing

College of Medical, Veterinary and Life Sciences

University of Glasgow

July 2023

Table of Contents

List of Tables

Chapter 1: Systematic Review

Chapter 2: Major Research Project

List of Figures

Chapter 1: Systematic Review

Chapter 2: Major Research Project

Acknowledgements

This project was a team effort. Thank you to my original supervisory team, Dr Naomi White, Dr Susan Cassidy & Dr Colette Montgomery Sardar for bringing the project to life. Naomi, your knowledge and passion for qualitative research was infectious, thank you for bolstering my confidence to push my interpretations further. A further thank you to Dr Jala Rizeq, your advisory support was pivotal to recruitment. The systematic review wouldn't have come to fruition without the expertise of Paul Cannon, and IT support from Blair Thompson, when technical difficulties threatened it all. Thank you to Professor Hamish McLeod & Dr Ruth Pegler for joining the supervisory team and supporting me to the finish line.

Thank you also to Dr Jenny Mclean, my clinical supervisor over the most intense phase of this project; your flexibility and encouragement provided much needed containment throughout. The same goes for all my Liaison colleagues, your fun and humour was much needed in this final year of training.

The research was only possible because of the Clinical Nurse Specialists within the Cardiology Service who persisted with recruitment, and of course the mothers who shared their infant's stories. Thank you for trusting me with those. I will remember fondly your abundant love and pride of your little ones. I hope this is a testament to my commitment to working in paediatric settings going forwards.

Thank you to all my friends and family for their love and encouragement during this long journey, I am blessed to have so many wonderful people around me. A special mention of my adorable nieces and nephews, some of whom were born in the midst of training. My appreciation to my fellow DClin peers. Especially to Kirsten as my second reviewer and to Sharena for being my study buddy and helping me find my way when I was lost amongst the themes.

To my best friend, Beth, who inspired me to get into this career in the first place. Thank you for always being my biggest cheerleader in anything I do.

To our pup, Winnie, thank you for remaining by my side (or feet) during the many hours spent at my desk and being the best reminder to stop and take a break.

Lastly, to my fiancée, Anton. Your patience and understanding knows no bounds and I'm not sure I'll ever be able to express how grateful I am to have you. My safe base always.

Agora, vamos casar!

Chapter 1: Systematic Review

Children and Young People’s Experiences of Living with Inflammatory Bowel Disease: A Systematic Review and Thematic Synthesis

Prepared in accordance with the author requirements for:

BMC Psychology

https://bmcpsychology.biomedcentral.com/submission-guidelines/preparing-your-manuscript

Abstract

Inflammatory Bowel Disease (IBD) refers predominantly to two chronic conditions that cause inflammation in the intestinal tract, Ulcerative Colitis and Crohn's Disease. The disease involves periods of 'flare-ups,' whereby symptoms significantly worsen, and then periods of remission. The number of children and young people (CYP) diagnosed with IBD is increasing; therefore, developing a deeper understanding of their experiences of living with this disease is imperative.

Objective

This review aimed to systematically identify, critically appraise, and thematically synthesise qualitative research exploring CYP's experiences of IBD.

Methods

An electronic search of five databases identified nine eligible articles. The Joanna Briggs Institute Critical Appraisal Checklist for Qualitative Research was used to assess the methodological quality of articles. Thematic Synthesis was used to generate analytic themes.

Results

The quality of studies was diverse, with some appraised as lower quality and lacking important methodological information. An overarching theme was developed: *An Abrupt Diversion*, reflecting the inherently disrupting nature of IBD upon CYP lives, including how they relate to themselves, others around them, and vice-versa. Themes within this overarching concept included: *An Altered Sense of Self; The Disclosure Decision: an Illusion?; and Allies in Battle*.

Conclusions

IBD itself, and/or other people's responses to it, altered CYP's identity and aspects of their development. Experiences appeared to vary over time and disease status; however, limited contextual information hindered further interpretations. Nuanced differences between child and adult IBD populations were found, which have implications for those around CYP in terms of supporting adjustment to IBD.

Keywords: inflammatory bowel disease, children, young people, adolescents, experiences, perspectives.

Introduction

Inflammatory Bowel Disease (IBD) refers predominantly to two chronic conditions that cause inflammation in the intestinal tract (Ulcerative Colitis [UC] and Crohn’s Disease [CD]; NHS, 2020). Common symptoms include greater frequency/urgency of bowel movements, diarrhoea, weight loss, stomach pain, anal bleeding, fatigue, or even growth failure (Raza et al., 2019). Individuals experience ‘flare-ups’, whereby symptoms significantly worsen, followed by periods of remission. Treatment options vary depending on stage and severity of disease; however, many have negative side effects. Options include medications, lifestyle adaptations, dietary restrictions, and/or routine endoscopic procedures. One in five people with severe UC and between 60-75% with severe CD will require surgery (NHS, 2020). Psychosocial impacts of IBD are extensive, including reduced quality of life, emotional distress, increased mental health (MH) difficulties, and poorer interpersonal relationships (Knowles et al., 2018a). The emotional consequences of IBD can exacerbate symptoms, creating a vicious cycle between physical and MH difficulties (Sun et al., 2019).

Prevalence of IBD in Children and Young People (CYP)

An estimated 8000 CYP in the UK live with IBD (IBD UK, 2019), reflecting the gradual rise in incidence worldwide, most notably in areas with previously low incidence (Cosnes et al., 2011). Although causes are unclear, researchers implicate factors such as diet, lifestyle, and environmental changes (Ponder & Long, 2013). CYP are usually diagnosed in adolescence, presenting with atypical symptoms that delay recognition, alongside a lack of gold standard testing procedures for this population (Griffiths, 2004). Diagnostic inequalities have been found, with African-American or Black CYP commonly experiencing delayed diagnosis (Barnes et al., 2021), increasing preventable disease complications and/or quicker progression to more severe symptoms (McLoughlin et al., 2021). Rising prevalence rates of IBD, alongside its early onset and incurable nature, has considerable cost implications for the healthcare system (The Lancet, 2012).

CYP with IBD

Psychosocial impacts of IBD for CYP are wide ranging and common. For example, attending school less frequently, academic-related difficulties, social difficulties, poorer self-esteem, and comorbid depression or anxiety disorders (Barnes et al., 2020; MacKner et al., 2006). Risk factors for psychosocial comorbidity in CYP include lower socioeconomic status, diagnosis in adolescence, greater disease severity, steroid treatments, and parental stress (Brooks et al., 2016). Existing systematic reviews (SR's) regarding CYP with IBD have, to date, been of quantitative design only, with narrow foci, such as prevalence of sleep disturbances (Moorman et al., 2023), efficacy of self-management interventions (Trans & Mulligan, 2019), or exploration of protective factors for psychosocial coping (Tempchin et al., 2021). A two-part quantitative SR explored quality of life in CYP and adult IBD populations (Knowles et al., 2018a; 2018b), and found poorer quality of life compared to healthy controls across the lifespan, although study quality was modest. Within-group differences of IBD were explored and poorer quality of life during active disease stage was found, as well as improvements in quality of life over longer disease duration, therefore suggesting adjustment over time in some individuals. A limitation of this review is that the included studies related disproportionally to the adult population rather than CYP with IBD (e.g. less than a quarter of total studies related to CYP in each review). Knowles et al (2018b) also recommended that future research explores potential factors related to quality of life, which qualitative explorations of overall IBD experience may be well-positioned to achieve.

Current Understanding of the Experiences of Living with IBD

The priorities and needs of those with IBD often differ from those of health care professionals (HCPs) treating them (Cervesi et al., 2013), meaning holistic, high quality and personalised care is not always achieved, despite ambitions from IBD UK (2019). Qualitative evidence synthesis is well positioned to enrich understandings of the needs and experiences of those with IBD. Prior SR's have explored experiences of living with IBD more generally and include qualitative studies from individuals with IBD across the lifespan. For instance, a SR by Fourie et al. (2018) highlighted the numerous challenges those with IBD face, evidenced by an overall theme *'feeling isolated and excluded'*. A SR by Muse et al. (2021) similarly reported an overarching theme *'feelings of otherness'*, influenced by personal, interpersonal and social factors. Although similar to the present review's focus, these prior reviews only included a small number of studies relating to CYP and did not report separate findings for the different age groups. CYP with IBD differ from their adult counterparts in terms of experience in a number of ways, including likely having a recent diagnosis, different disease courses, being seen by a separate paediatric service, all alongside experiencing numerous developmental challenges; therefore, drawing upon combined adult-child findings is not sufficient nor appropriate to fully understand the experiences and needs of CYP with IBD. The current review seeks to address this gap in the IBD literature by focusing exclusively upon understanding CYP’s experiences of IBD.

Review Rationale

CYP’s experiences of IBD remains under-researched and the quality of the evidence base is not established. CYP experience many developmental challenges, for example: developing their independence/autonomy, deepening friendships, exploring sexuality, and identity consolidation, which may be compounded or complicated further by IBD. Thus, drawing upon combined findings from child and adult populations is not sufficient to fully understand CYP’s unique experiences. Moreover, there is likely to be variation between CYP with IBD, according to their respective developmental stages, and/or across different sociocultural contexts. Although there is extensive quantitative literature available describing disease-specific management issues, as outlined above, this neglects the patient's voice and overlooks their common broader experiences of living with this disease.

Aims

The aim of this review was to systematically identify, critically appraise, and thematically synthesise the available qualitative research exploring CYP’s experiences of IBD. It is anticipated that this review, generating novel insights, will enhance theoretical understandings of the experiences of CYP with IBD and may inform service developments for paediatric patients.

Review Question

What are CYP’s experiences of living with IBD?

Methods

Design

This was a qualitative SR and a PROSPERO protocol was registered on 14th June 2022 (registration number: CRD42022339004). This review followed qualitative reporting guidelines, i.e. ENTREQ (Tong et al., 2012; see Appendix A, p. 107).

Inclusion Criteria

CYP with a diagnosis of IBD; peer-reviewed research using qualitative methodology (Flemming et al., 2018), or mixed-methods research provided qualitative data could be extracted separately; experiences of living with IBD, including perceptions, perspectives, or considerations of IBD; no restrictions were implemented on location or setting (inpatient/outpatient); studies published in full and of English language.

Exclusion Criteria

Quantitative studies or those focused on the transition to adult services (see existing review: Clarke & Lusher, 2016); adult participants where it was not possible to extrapolate separate findings from CYP, i.e. those reporting combined findings from CYP-parent or CYP-clinician; studies of IBD interventions, rather than focusing on broader experiences of living with IBD; studies reporting on multiple physical health conditions.

Search Strategy and Terms

PICo (Population, Phenomena of Interest, & Context) framework guided the overarching search concepts (Munn et al., 2018):

- **Population**: CYP with IBD
- **Phenomena of Interest**: Experiences of living with IBD
- **Context**: No restrictions on location of study or setting

The search strategy combined three key concepts from relevant, peer-reviewed SRs to enhance validity (Tran & Mulligan, 2019; Shaw et al., 2004). Specifically, Tran and Mulligan's (2019) 'IBD' and 'CYP' search terms, alongside search terms for 'qualitative research' by Shaw et al. (2004). The search used a combination of thesaurus/index terms (translated for each database) and free text words. Search terms were combined using Boolean operators "or" / "and". No date restrictions were imposed given this was an original SR. This strategy was developed in collaboration with the University's Specialist Librarian. The final search was carried out on 27th June 2022, see Appendix B for examples of the search strategy (p. 108).

Database Searches

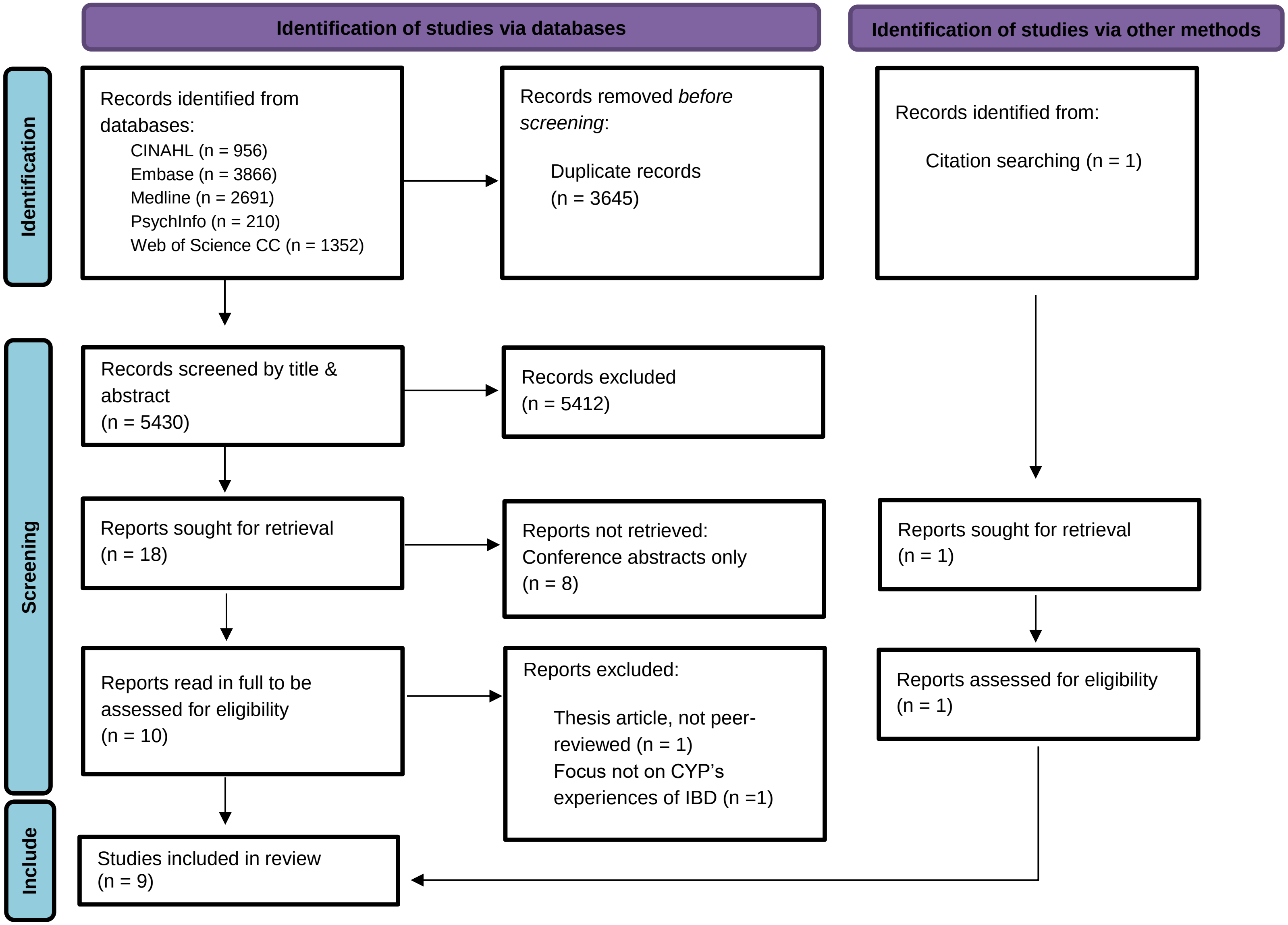
A scoping search, including Prospero and Epistemonikos, revealed no existing or ongoing SRs specific to CYP's experiences of IBD. Five electronic databases across psychology, gastroenterology, and nursing were purposively selected to retrieve relevant articles (CINAHL, Embase, Web of Science Core Collection, PsychINFO and Medline). Forward and backward citation searches of eligible papers were conducted using Google Scholar.

Study Selection

Database searches identified 9075 records, which were imported to EndNote. 3645 duplicates were removed. The remaining 5430 records were title and abstract screened by the lead reviewer. A second reviewer (a Trainee Clinical Psychologist) screened a subset to ensure accuracy and no disparities were found between reviewers (all articles retrieved from Medline). 5412 records were excluded, meaning 18 records remained for full-text review. Eight records were excluded as these were conference abstracts only and did not contain the author's contact details to request full text. One record was excluded because it was not peer-reviewed, and another as it did not relate to IBD experiences. Three of the remaining eight records included a small proportion of participants aged 19 years old. Although UK paediatric services typically only include CYP aged up to 18 years old, no studies originated from the UK and the World Health Organisation (2016) defines adolescence as up to 19 years old. Participants were all recruited from paediatric services, with mean age below 18 years. Excluding these articles would have narrowed this review's breadth and lost valuable data, coherent to the RQ. Backwards and forwards citation searching found one additional study that met eligibility criteria. Figure 1 provides an overview of this process.

Figure 1

PRISMA Flow Diagram



Data Extraction

Data were extracted using a standardised extraction format on Microsoft Excel (see Table 1). The second reviewer cross-checked four of the included articles to ensure data had been extracted accurately.

Critical Appraisal

The myriad of quality appraisal tools available for qualitative research reflects lack of consensus regarding what constitutes methodological ‘quality’ in this field. Within this review, studies were not excluded based on their quality rating; rather, less emphasis was given within the synthesis to findings from studies assessed as relatively lacking in methodological rigour. The Joanna Briggs Institute (JBI) Critical Appraisal Checklist for Qualitative Research was utilised to assess all articles (Lockwood et al., 2015; see Appendix C, p. 118), given it is sensitive to interpretative and theoretical validity (Hannes et al., 2010). The JBI has 10 items and responses included ‘yes’, ‘no’, ‘unclear’ or ‘not applicable’. The second reviewer appraised four of the nine (44.4%) eligible studies, selected at random. Initial inter-rater agreement was 95% (38/40); the common difference in initial opinion was whether researcher influence on the research was sufficiently addressed or not. Disagreements were discussed, with guidance from academic supervisor, until consensus was reached for all items.

Data Synthesis

Thematic Synthesis (TS; Thomas & Harden, 2008) was the chosen analysis method. TS is the method used predominantly for qualitative SRs focusing on individual's experiences of a topic and is advantageous for informing practice (a secondary aim of our review; Carroll, 2017). TS allows for heterogeneous studies to be synthesised, which was necessary in the current review given that the included articles used diverse analytic approaches (i.e. thematic approaches, idiographic approaches, ethnography, etc). Each study's analytic choice/method did not directly inform synthesis; rather, the extent of adherence with their reported approach and interpretative depth were more relevant quality indicators for synthesis contribution.

TS is an accessible option of synthesis for novice reviewers and was appropriate to this review's time constraints. TS allows for new understandings and inferences to be developed, whilst also preserving conclusions from primary studies. Although TS does not explicitly align itself with a particular epistemological stance, it does to some extent assume that its output, involving some level of interpretation, reflects a shared reality, mediated by participants', authors', and reviewer's beliefs/perceptions (Barnett-Page & Thomas, 2009), aligning with the lead reviewer's critical realist stance.

Synthesis followed three stages. Stage 1 involved coding studies line by line; coding was therefore inductive and number of codes increased as coding progressed. All information provided within the results section and any relevant portions or conclusions drawn in the discussion section were considered as data and coded. Data therefore comprised of a combination of the author's interpretations and participants direct quotes. Stage 2 involved clustering related codes within a study to create 'sets', which were then compared and collated across studies to develop descriptive themes. This required staying ‘close’ to original texts, whilst also translating key ideas into broader patterns of meaning. The original studies were frequently revisited to ensure developing themes were in keeping with key findings. In stage 3, more abstract meanings from descriptive themes were generated in the form of analytic themes. Analysis was predominantly undertaken by the lead reviewer, in ongoing consultation with academic supervisors.

Although authors' summaries and interpretative statements were important data that contributed to the synthesis process, only direct participant quotes were included in the synthesis write up. The decision to include only direct participant quotations, as opposed to author quotations, was two-fold: first, to demonstrate transparency and that reviewer's interpretations (e.g. analytic themes) were grounded in the original papers; secondly, to ensure CYP’s voices and their experiences were sufficiently represented throughout, a key aim of this review.

Reflexivity Statement

The lead reviewer was a Trainee Clinical Psychologist who had worked in various Child and Adolescent MH teams to support CYP with a range of MH difficulties. With no personal experience and limited clinical experience of working with CYP with IBD, the lead reviewer had a predominantly ‘outsider’ perspective of IBD. Supervision was used to reflect on any pre-existing assumptions, beliefs, and feelings from prior clinical experiences, in aim of increasing reflexivity whilst undertaking TS, as well as developing a reflective log. This review was carried out concurrently with a qualitative research project, also adopting a critical realist stance; thus, the author’s guiding epistemological stance will, to some extent, have influenced meaning and sense derived from data in this review (see Appendix D, p. 119).

Results

Study Characteristics

Table 1 presents an overview of study characteristics. The nine studies were published from 1996-2022, and were undertaken in three countries: Sweden, Denmark & Canada. Studies explored various aspects of living with IBD, including broad impacts on daily living[1,2,4,7], strategies and supports utilised to cope[2,6,7], disclosure decisions about IBD[3], how others respond to IBD[5,6], experiences of hospitalisation for surgery[8], and experiences of living with an ostomy[9].

In terms of design, one study was mixed methods[4], whereas the rest were qualitative. Data were predominantly collected from individual semi-structured interviews with participants. Exceptions to this included one study that allowed a proportion of the sample to provide written responses rather than participate in an interview[6]; the mixed methods study[4] that captured data from open-ended questions on a questionnaire; and another[9] that used a focus group after individual interviews. Three studies employed content analysis[4,7,9]; two used grounded theory[5,6]; one used thematic analysis[2]; one used a phenomenological hermeneutical method[1]; one used an unspecified 'hermeneutic' method[8]; and one study did not specify analysis method[3].

Across studies, there were 244 participants in total, all of whom had IBD. Two articles came from the same sample[2,3], focusing on distinct experiences of IBD, and so these participants were only counted once. Studies[7,9] recruited from the same hospital as part of a larger intervention-based study, meaning it is possible that there was some overlap in the samples in these studies. However, as this could not be confirmed, both samples were included separately within overall numbers. All participants were outpatients at a children's hospital at the time of study participation; however, at least a proportion of participants had been inpatients in relation to IBD at some point[8,9]. One study did not specify participants' IBD diagnosis[9]; however, from the studies that did, 114 participants (50.89%) had UC; 107 (47.77%) had CD; and 3 (1.34%) had indeterminate colitis. Most articles did not specify severity of participants IBD; however, across the articles included there was variation of IBD severity, given some participants were hospitalised for IBD and required stomas. There were 99 (44.20%) female participants and 125 (55.80%) males. The sample included participants aged 7-19 years old. The youngest mean age reported in any given sample was 13.3 years[7] (study[1] reported median age of sample rather than mean), and the oldest mean age was 16.5 years[8]. No studies provided details of participants' ethnicity; however, three studies[3,7,9] provided qualitative descriptors of their sample as "diverse".

Table 1

Study Characteristics and Findings

Reference **Study No.** **Country (Setting/ Context of study)**	**Aims**	**Sample Size (Age, gender, & ethnicity)**	**IBD Diagnoses (Characteristics of condition, where stated)**	**Philosophical Perspective & Methodology**	**Methods of Data Collection & Analysis**	**Key Findings**
Vejzovic et al. (2018) [1] Sweden (Outpatients at Paediatric Hospital)	Illuminate the meaning of children's lived experiences of IBD.	N=7 ○ 13-18 years (Median= 15) ○ 4 males, 3 females ○ Ethnicity not specified	100% UC ○ Duration of illness: 1–3 years	Phenomenological Hermeneutics	Semi-structured interviews, analysed using "Phenomenological Hermeneutical" method	Main theme: "A daily struggle to adapt and to be perceived as normal" Four subthemes: 1. Being healthy despite the symptoms 2. Being healthy despite being afraid

						3. Being healthy despite a sense of being different 4. Being healthy despite needing support.
Barned et al. (2022) [2] Canada (Outpatients at Children's Hospital; Additional info: free healthcare, hospital in area of high median incomes, & high levels of education).	Explore illness experiences of CYP with IBD to discern what issues they face, and what lifestyle adjustments and strategies they devise in response to their circumstances.	N=25 ○ 10-17 years (Mean= 13.8) ○ 13 males, 12 females ○ Ethnicity not specified	32% UC 68% CD ○ Reported variation in severity, treatment type, and age at diagnosis	Neither stated	Semi-structured interviews, analysed using Thematic Analysis	<u>Three themes:</u> 1. Challenges related to diagnosis 2. Managing identity and making sense of change 3. Navigating sociability.

Barned et al. (2016) [3] Canada (Outpatients at Children's Hospital; Additional info: free healthcare, hospital in area of high median incomes, & high levels of education).	Perspectives of Canadian children and adolescents living with IBD to determine how they go about deciding if and when to tell others about their illness.	N=25 ○ 10-17 years (Mean= 13.8) ○ 13 males, 12 females ○ Ethnicity not specified	32% UC 68% CD ○ Reported variation in severity, treatment type, and age at diagnosis	Neither stated	Semi-structured interviews, analysis method not explicitly stated. ○ Does refer to re-reading transcripts in order to "identify consistent themes"	Major theme: Illness disclosure important to understanding children and adolescents' illness experiences. Three key factors: 1. To disclose or conceal: making the decision 2. When to tell: factors influencing disclosure decisions 3. Challenges of IBD disclosure: the reactions of others.
Lindfred et al (2012) [4]	Explore and describe	N=67	63% UC 33% CD	Neither stated	Mixed-methods design, qualitative	Three headings: 1. Symptoms & Health Habits

Sweden (Outpatients at Paediatric Hospital)	adolescents' perceptions of health, self-management, and the impact of living with IBD in daily life.	○ 11-16 years (Mean= 14.6) ○ 40 males, 27 females ○ Ethnicity not specified	4% IC ○ Mean duration of illness: 2.9 years ○ 37% mild IBD, 28% moderate IBD, 34% severe IBD ○ 31% in relapse at time of interview, 68% in remission		data from open-ended questions on questionnaire, analysed using “manifest content analysis”	2. Knowledge, Responsibility, and Communication with Parents 3. Self-Image and the Impact of IBD on Daily Life
Reichenberg et al. (2007) [5]	Identify how adolescents with IBD respond to	N=17 ○ 12-18 years (Median/Mean = Not stated)	59% UC 41% CD	Not stated (constructionist perspective implied but not	Semi-structured interviews, analysed using Grounded Theory	Main theme: Ambivalence Three subthemes: 1. Ability/inability

Sweden (Outpatients at Paediatric Hospital)	their parents' concern for them	○ 10 males, 7 females ○ Ethnicity not specified	○ Duration of illness: 1–5 years ○ 18% mild IBD, 29% moderate IBD, severe IBD 53%	stated) & Grounded Theory		2. Compliance/resistance 3. Trust/distrust.
Holaday et al. (1996) [6] Sweden (Outpatients)	Describe what behaviour from the environment was perceived and valued as supportive by adolescents with IBD.	N=20 ○ 11-19 years (Mean= 14.8) ○ Gender not specified ○ Ethnicity not specified	100% UC ○ 75% had had surgery for UC	Not stated & Grounded Theory	Interviews conducted with 15 participants & 5 provided written responses to interview questions, analysed using “constant comparative method” from	<u>Five categories:</u> 1. Additional material support 2. Situational humanistic support 3. Professional support with 2 dimensions (instrumental support and emotional support) 4. Affiliational support 5. Emotional support with 3 dimensions (emotional

					Grounded Theory	advisory support, emotional safety support, and self-ideal emotional support)
Nicholas et al. (2007) [7] Canada (Outpatients at Children's Hospital)	Understand the lived experience and elements of quality of life as depicted by children and adolescents with IBD.	N=80 ○ 7-19 years (Mean= 13.3) ○ 44 males, 36 females ○ Descriptor: "ethnically diverse sample"	24% UC 76% CD ○ Minimum duration of diagnosis: 6 months	Interpretative approach & Ethnography	In-depth interviews, analysed using Content Analysis	Common experiences: 1. Concerns relating to IBD symptoms and treatments 2. Vulnerability and lack of control 3. Perceiving the self negatively and as different than peers 4. Benefits of social support 5. Personal resources in coping "While the experiences described were not universal for all participants it was typically found that children with more severe symptoms were more likely to

						express greater psychosocial difficulty.”
Olsen et al (2016) [8] Denmark (Outpatients, prior hospitalisation for surgery)	Identify and describe adolescents’ lived experiences while they were hospitalized after surgery for UC.	N=8 ○ 13-19 years (Mean= 16.5) ○ 5 males, 3 females ○ Ethnicity not specified	100% UC ○ Mean duration of illness: 3.6 years ○ Interviewed 2-5 months after discharged home post-surgery	Not stated & Hermeneutic Interpretation*	Semi-structured interviews, analysed using “Hermeneutic interpretation of meaning”	<u>Three themes:</u> 1. Body: Out of Order 2. Seen and Understood 3. Where are all the others?
Nicholas et al (2008) [9] Canada (Outpatients at Paediatric Hospital)	Explore adolescents' experiences and quality of life following ostomy surgery.	N=20 1. 13-19 years (Mean= 15.3) 2. 9 males, 11 females 3. Descriptor: “culturally	Doesn’t specify type of IBD diagnoses ○ Mean time since surgery: 2.9 years (ranging from	Interpretative approach & Ethnography	Semi-structured interviews, followed by a focus group with same participants, analysed using Content Analysis	<u>Seven themes:</u> 1. Body intrusion and body image (BI) challenges 2. Decreased independence and control 3. Secrecy: Considerations in deciding whether to tell others about the ostomy

	diverse participants”	3 months – 10 years)		4. Adjustment over time 5. Challenges for the family 6. Sources of strength: Family and friends 7. Benefits and growth

Critical Appraisal

The quality of studies was diverse, with a few papers of higher quality and others that were lower (see Table 2 for an overview). Overall, no studies fulfilled all criteria appraised (i.e. a response of 'yes' to all 10 questions), with a minimum rating of 2/10 criteria fulfilled[4] and a maximum of 9/10 fulfilled[1]. Only three studies included a statement explicitly stating their philosophical perspective or theoretical premise[1,7,9]. One study seemed to imply a constructionist perspective but did not make this explicit[5]. The remaining five studies[2,3,4,6,8] did not make any reference to such, making it impossible to determine congruence with study methodology thereafter.

Inclusion of a clear methodology was similarly inconsistent, likely reflecting ambiguity in the qualitative field as to what constitutes 'methodology' and how this is distinct from 'methods' and philosophical perspective. Again, this had implications for reviewer's ability to determine congruence with other study features thereafter (e.g. questions 2-5 on JBI appraisal tool). Although all studies included clear research questions, in two studies these were somewhat incongruent with subsequent methodology. For instance, studies[6,8] stated their aims were to "describe" certain experiences of IBD; however, then used methodologies that emphasise interpretation as opposed to mere description. Furthermore, it was unclear from their descriptions of analytic process the extent that they adhered to principles of hermeneutics and GT referenced. In studies clearly stating their methodology, data collection methods were congruent with this[1,5,6,7,8,9].

Most notably absent from studies was a statement locating the researcher culturally or theoretically, as well as a lack of information about researcher influence on the research (and vice versa). This may have been omitted due to journal word counts or could reflect limited reflexivity throughout the research process. Regardless, such omissions made it more challenging to determine credibility and validity of the subsequent reported findings.

A strength of the studies was the representation of participants' voices, with almost all studies including sufficient quotes to adequately demonstrate and/or corroborate authors' findings and interpretations. The exception to this was study[4] who only included a few brief quotes, perhaps because of its mixed-methods design. Two studies[6,8] did not indicate they obtained appropriate ethical approval for the research, although study[8] did reference “following appropriate ethical guidelines”.

Table 2

Critical Appraisal using JBI Checklist

	Q1	Q2	Q3	Q4.	Q5.	Q6	Q7	Q8	Q9	Q10
	Congruence between methodology and…					Information on researcher cultural / theoretical stance	Researcher reflexivity	Adequate representation of participants voices	Evidenced ethical approval	Appropriate/ relevant conclusions
Authors (Publication Year)	Philosophical perspective	Research question (s)	Methods	Data analysis & representation	Interpretation of results					
Vjzovic et al (2018)*	Y	Y	Y	Y	Y	N	Y	Y	Y	Y
Nicholas et al (2007)*	Y	Y	Y	Y	Y	N	N	Y	Y	Y
Nicholas et al (2008)	Y	Y	Y	Y	Y	N	N	Y	Y	Y
Barned et al (2016)*	U	U	U	U	U	N	N	Y	Y	Y
Barned et al (2022)*	U	U	U	U	U	U	N	Y	Y	Y
Olsen et al (2016)	U	U	Y	Y	Y	N	N	Y	U	Y
Reichenberg et al (2007)*	U	Y	Y	Y	Y	N	U	Y	Y	Y

Holaday et al (1996)	U	N	Y	N	Y	N	N	Y	N	Y
Linfred et al (2012)	U	U	U	U	U	N	N	Y	Y	U

Yes = Green; Unclear = Amber; No = Red; Not Applicable = Blue.

‘No’ ratings were given when a study specified relevant information related to the item but demonstrated incongruence.

‘Unclear’ ratings were given when a study did not include the necessary information to determine congruence.

Studies were not excluded on the basis of quality, as methodologically weaker studies could still provide meaningful, rich insights relevant to the RQ of this review.

Rather, methodologically weaker studies or those with less interpretative depth contributed less so to analytic theme development during thematic synthesis.

Thematic Synthesis (TS)

TS generated one overarching analytic theme *“An Abrupt Diversion*”, alongside three themes and four subthemes, shown in Table 3 and discussed further below. Synthesis demonstrated IBD alters how CYP relate to themselves, as well as how they relate to their network and vice versa. The distribution of themes across articles can be found in Table 4; themes were not present in all studies as each study related to different aspects of living with IBD, some with narrower focuses than others. A coding extract can be found in Appendix E (p. 120).

Table 3

TS Findings

Overarching Theme*: An Abrupt Diversion*		
Descriptive Themes	Analytic Themes	Subthemes
Self to Self Relating	*Theme 1: An Altered Sense of Self*	a) *“Disease?! What?”: Disbelief and Denial*
		b) IBD as an Unwanted Intruder
		c) Discovering Strengths in the Struggle
Self in Relation to Others	*Theme 2: The Disclosure Decision: An Illusion?*	
	Theme 3: Allies in Battle	a) *A Diminished Self*

Table 4

Theme Distribution Table

	Themes and Sub-Themes							
		Self-Self Relating				Self in Relation to Others		
Reference	Overarching theme: An Abrupt Diversion	Theme 1: An Altered Sense of Self	Subtheme 1a: “Disease?! What?”: Disbelief and Denial	Subtheme 1b: IBD as an Unwanted Intruder	Subtheme 1c: Discovering Strengths in the Struggle	Theme 2: Disclosure Decision: An Illusion?	Theme 3: Allies in Battle	Subtheme 3a: A Diminished Self
1. Vejzovic et al (2018)	Y	Y	Y	Y	Y	Y	Y	Y
2. Nicholas et al (2007)	Y	Y	Y	Y	Y	Y	Y	Y
3. Nicholas et al (2008)	Y	Y	**N**	Y	Y	Y	Y	Y
4. Barned et al (2016)	Y	Y	Y	Y	**N**	Y	Y	Y
5. Barned et al (2022)	Y	Y	Y	Y	Y	**N**	**N**	**N**
6. Olsen et al (2016)	Y	Y	**N**	Y	Y	**N**	Y	Y

7. Reichenberg et al (2007)	Y	Y	N	Y	N	N	Y	Y
8. Holaday et al (1996)	Y	Y	N	N	N	Y	Y	Y
9. Linfred et al (2012)	Y	Y	Y	Y	N	N	Y	Y

Y = Yes; N = No

Overarching Theme: An Abrupt Diversion

Themes were nested within the overarching idea of IBD as an *'An Abrupt Diversion'* to CYP's lives. Across studies, participants explicitly and, or implicitly portrayed the diagnosis itself, and then periods of disease exacerbation thereafter, as halting their journey of childhood, adolescence, and development. For example: *"It [IBD] has stopped my life"* (Nicholas et al., 2007, p.4). As this quote denotes, IBD symptoms and treatments had an inherently disrupting influence upon all aspects of CYP's daily lives, including their identity, body image (BI), how they interact with others and the world around them, and vice-versa. A loss of control was depicted, with IBD symbolised as the roadblock that CYP had to unexpectedly navigate. Authors suggested the extent of diversion correlated somewhat with disease stage and/or severity; however, few articles contextualized findings or quotations with participant disease severity, so this could not be clearly discerned.

IBD altered CYP's ability to engage in meaningful activities in their lives, including attending school and socialising with peers, all representing losses from what the CYP had imagined for this stage of life. As one participant described: *"…the rest of my childhood was lost"* (Barned et al., 2022, p. 99). Common across participants' quotes was a blunt use of language, reflecting the abrupt variations to their lives. CYP made comparisons to their trajectories pre-IBD, as well as to their peer's lives, and felt isolated in their new normality; for example: *"I'm not a part of it [socialising]. They're [friends] getting on with their lives, like boyfriends and parties, and I'm going through a horrible time"* (Nicholas et al., 2007, p.5), also evidenced by the overarching finding in Vejzovic et al. (2018, p.335): *“A daily struggle to adapt and to be perceived as normal”.*

Theme 1: An Altered Sense of Self

IBD altered CYP’s sense of self in multiple ways, permeating physical, cognitive, perceptual, affective, and social aspects of self. The diagnosis had significant implications in terms of CYP’s identity; the symptoms and/or treatments had a derailing effect upon one’s ‘expected’ course of development; and CYP’s BI and physicality became distorted. In five studies, acceptance and post-diagnostic growth were referenced, whereby IBD was integrated within their sense of self; however, this was only a facet of singular themes as opposed to being an overarching finding.

Subtheme 1a: “Disease?! What?”: Disbelief and Denial

Participants described overwhelming emotional responses to receiving a diagnosis, including high levels of uncertainty about what this would mean long-term. What appeared prominent throughout the analytic process, was a struggle to reconcile being both a CYP and someone with a “disease”, creating dissonance between their preconceived ideas and current circumstances. Participants voiced confusion, disbelief, and then, attempts to deny such a label, as evidenced in the following quote:

> *“...when you think disease, you think like a sick person in a hospital bed struggling for their life kind of thing... So to me, when they say, “Oh you have Crohn’s Disease”, I’m like, “Disease?! What?””* (Barned et al., 2022, p.100)

One participant struggled to even verbalise “disease” aloud (Barned et al., 2022), whilst another alluded to it as something to be ashamed of (Barned et al., 2016). Narratives suggested some level of disavowal, likely reflecting unintentional coping strategies. CYP found it difficult to integrate IBD initially within their overall sense of self and there was a sense of being permanently changed thereafter.

Subtheme 1b: IBD as an Intruder

Across studies, participants conveyed IBD as an unwanted presence, invading their body internally via symptoms and/or treatments, and externally, by altering their BI and appearance.

> *“You actually don’t know what is going on inside your body”* (Olsen et al., 2016, p.289)

> *“The prednisolone made me look like a meatball”* (Lindfred et al., 2012. p.260)

Self-criticism was entrenched within CYP’s descriptions, provoking disgust and/or shame, despite having little control over the impacts of IBD. In addition to disturbed BI, the boundaries of CYP’s bodies as their own seemed to blur as a result of IBD. The CYP described others as intruding their physical space and their bodies because of IBD; for example, when discussing HCPs, one YP described:

> *“She [HCP] was very domineering … ‘Now we do this’ and ‘Now we do that’, you know”* (Olsen et al., 2016, p.291)

> *“My Mum comes by when I’m on the toilet to see what is wrong.... It’s uncomfortable to have my mum watch me on the toilet”* (Nicholas et al., 2007, p4).

IBD itself, or other people's responses to it, was personified in the data as a constant, punitive, unwelcomed presence, intruding upon CYP's sense of self; for example: *"It [IBD medication] makes me have this stomach"* (Barned et al., 2022, p. 100).

Subtheme 1c: Discovering Strengths in the Struggle

Whilst acknowledging it had been a complex process to get to this point, a small minority of participants had come to accept their new path with IBD, and in some instances, had realised personal strengths they felt they wouldn't have otherwise. For example:

> *"I've accepted that things won't return to normal, even if there are moments when it feels as if it is"* (Vejzovic, et al., 2018, p. 336)
>
> *"I'm a gentler person now"* (Nicholas et al., 2007, p.6).

Participants conveyed a sense of gratitude, from feeling enriched psychologically by the challenges and struggles they had endured. These CYP demonstrated a commitment to moving forwards with IBD, whilst acknowledging the disease's unpredictability. As one participant conveyed when describing living with an ostomy: *"Leaks will happen […]. Sometimes you go for years without leaks, sometimes every other day you'll have a leak. It's not so bad. Clean up, change, done."* (Nicholas et al., 2008, p.5). Participants reported increased resilience and confidence in their ability to cope with future potential adversities, illustrating a sense of mastery and achievement from IBD challenges already overcome. This adjustment process likely varied significantly between CYP in terms of timing and severity of disease; however, due to limited demographic information accompanying findings it was not possible to discern any shared characteristics of this subsample.

Theme 2: The Disclosure Decision: An Illusion?

Participants described deliberating at length about whether to disclose IBD to peers or not, deeming this a risky and complex decision to make. Although participants and authors predominantly spoke of this as a 'choice' or 'decision', often the profound impact of IBD upon daily life meant others were already aware of changes in the CYP, rendering CYP little choice but to tell others about their condition.

> *"Well they realized that there was something wrong because I didn't go out, I always stayed in because like with the stomach cramps and the diarrhoea, I didn't want to go out, so they [friends] realised, because I'm used to always being like the energetic one..... so they knew it was something and then I told them, like, there is something wrong with me"* (Barned et al., 2016, p.118-19)

Hence, the idea of a 'disclosure decision' was somewhat an illusion, or perhaps an attempt to regain some sense of control at a highly uncertain time. Disclosures were often made reluctantly, to prevent inaccurate speculation from peers (Vejzovic et al., 2018), or to explain changes in appearance or behaviour:

> *"...I didn't want to tell them at first but then they realized ok he's not here a lot of days and then he shows up with a bandage on his arm, so either he stays at home and stabs himself or he goes to the hospital"* (Barned et al., 2016, p. 117).

Thus, there was a sense that IBD can eradicate meaningful disclosure choice.

Theme 3: Allies in Battle

Participants situated their coping and adjustment to IBD within the extent and nature of support they received from others in their network. Their descriptions conveyed most caregivers and family members as allies in their battle who could empower and offer a safe base to make sense of IBD from. For example:

> *"My whole family stands up for me and I feel secure"* and *"The best support you can have from your parents is that they are just there […]. They feel your needs in the air"* (Holaday et al., 1996, p.75)

CYP valued being seen and supported as a whole person, beyond the disease, and retaining agency. The nature of the support received from others varied greatly amongst CYP, and at times these allies underestimated CYP's resilience, which led to a diminished sense of self, as described further below.

Subtheme 3a: A Diminished Self

Allies in the CYP's network could be experienced as over-bearing at times, diminishing aspects of their identity out with IBD, and stalling individuation, an important developmental task of this life stage. Although perceived by CYP as well-intentioned, over-bearing support often evoked feelings of incompetence and led to anxiety or shame from perceptions of burdening others. As one participant described: *"They ask you nearly every day how you're doing […]. They worry about every little thing – should we go to the hospital or something? That's what it's like."* (Reichenberg et al., 2007, p. 479), leading to hopelessness about life more generally and the future, *"everything is pointless"* (Nicholas et al., 2007, p.6). Participants from other studies voiced suicidal ideation, similarly stemming from guilt about the perceived toll of their IBD on others, *"I could sense that it was hard on my parents ... so maybe [dying] would just be the easiest thing"* (Olsen et al., 2016, p. 291)*.* This sense, or fear, of burdening others prevented CYP from fully sharing their own difficult thoughts and feelings (Vejzovic et al., 2018). For those that had been hospitalised, a similar pattern occurred in relation to nursing staff (Olsen et al., 2016). CYP demonstrated conflicting feelings of gratitude for parents or HCPs acting as allies where needed, alongside frustration as they desired some level of autonomy and agency. As level of involvement regarding CYP's IBD increased, it was as though their sense of self was diminished, until IBD became their core defining feature, within themselves, and as perceived by others.

Discussion

This review identified, appraised, and synthesised nine qualitative articles exploring CYP's experiences of living with IBD. Three themes and four sub-themes were developed, situated within an overarching concept: *An Abrupt Diversion.* This reflects the sudden and all-encompassing nature of how an IBD diagnosis disrupts CYP lives, shattering expectations and aspects of their identity and development. Sub-themes within an *Altered Sense of Self* reflect the various ways CYP's relationship with themselves is altered by IBD, including: *"Disease?! What?": Disbelief and Denial* at having to identify with the diagnosis; the portrayal of IBD as *an Unwanted Intruder* invading the CYP's body image (BI) and physical boundaries; and some CYP feeling enriched psychologically by IBD, evidenced by *Discovering Strengths in the Struggle. The Disclosure Decision: an Illusion?* illustrates how meaningful choice or privacy is diminished by symptoms of IBD and treatment side effects. Finally, *Allies in Battle,* and sub-theme *A Diminished Self,* depict the impact of other people's responses to

IBD upon how CYP feel about themselves, either resulting in empowerment or further vulnerability. Overall, these findings share commonalities with CYP's experiences of other chronic health conditions; specifically, psychological experiences of shame, avoidance, and identity overshadowing, alongside social implications such as lack of autonomy/privacy and disclosure concerns (Shorey & Ng, 2020).

The overarching theme within this review *'An Abrupt Diversion'* is somewhat comparable to findings from adult IBD studies, including a "feeling of otherness" and "living in isolation and exclusion" (Muse et al., 2021 and Fourie et al., 2018 respectively). Although broadly similar, subtle differences in the overarching concepts between the populations could be understood by the developmental implications of IBD. Erikson's Theory of Psychosocial Development (1968) postulates that identity formation is the pivotal task of adolescence, whereas adulthood is marked by conflict around intimacy versus isolation. Indeed, within this review IBD was experienced by CYP as markedly disrupting the developing self and identity, thus creating a crisis in this psychosocial stage of development; whereas findings from adults with IBD demonstrate greater concern about relating to others. Whilst relating to others was one facet of CYP's experience, an altered sense of self from IBD appeared prominent during analysis. Adult samples will have had greater opportunity to adjust to diagnosis and will likely have already navigated the psychosexual/identity issues characterising adolescence, particularly if they experienced later IBD onset or adult diagnoses. Other findings from the adult IBD literature mirror some of the remaining themes, including alterations to BI and a sense of IBD burdening others. Both aforementioned reviews included a small number of the CYP articles included within the current review (e.g.

Barned et al., 2016; Lindfred et al., 2012; Nicholas et al., 2007; Vejzovic et al., 2018), which may to some extent underpin similar findings.

Also different to findings from adult populations was the greater variability in CYP's psychological experiences of IBD, given a sub-set described growth because of IBD experiences and acceptance. Hence, *An Altered Sense of Self* encapsulates potential positive consequences of IBD, in addition to its adversities. It is plausible CYP's relationship with IBD oscillates according to developmental stage; however, as all studies reported combined findings from younger and older children/adolescents, potential developmental differences could not be explored. Sub-themes within an *Altered Sense of Self* resemble coping patterns proposed by grief theories (i.e. the DABDA model; Kubler-Ross & Kessler, 2005); as CYP with IBD journeyed through denial/anger following diagnosis, bargaining to maintain normality, and then, in some cases, developed acceptance of their condition.

Disclosure to peers was interpreted as a momentous experience for CYP, echoing findings from other chronic health conditions whereby disclosure was central to the overall illness experience (Woodgate et al., 2022). What may be distinct to IBD is the illusion of choice regarding disclosure, given the nature of symptoms or behavioural changes (weight loss, frequent/urgent trips to the toilet, or fatigue). The theme *Allies in Battle,* and subtheme *A Diminished Self,* highlight *how* support is communicated and received by CYP with IBD. Together, this theme and subtheme reflect findings from non-IBD adolescents, whereby caregivers fluctuate between being a source of support or a stressor (Camara et al., 2017). Thus, adolescent's support needs vary, further complicated in CYP with IBD by fluctuations in disease status, meaning caregivers must adapt accordingly. In comparison to other chronic health populations, parents of CYP with IBD are more likely to endorse high emotional distress (Plevinsky et al., 2018), which provides some insight as to why caregivers may interact in an overbearing/anxious manner.

Weaknesses of the Literature

There were several weaknesses within the articles in this review. Most studies did not provide specific information about disease severity, or whether participants were in remission or relapse at the time of participation, meaning it was not possible to consider the potential influence of this on their views, experiences, or attitudes towards IBD. This may have helped to discern variability regarding sense of self, with some CYP feeling enriched by IBD. The relevance of these findings across ethnic groups is unknown given the absence of ethnicity information in studies. Given recent research highlighted health inequities in relation to IBD diagnosis and treatment for patients from ethnic minorities, this is pertinent to consider (Barnes et al., 2021). Researcher reflexivity was lacking from most studies, which is particularly problematic when multiple articles were undertaken by the same lead author (i.e. Barned et al. and Nicholas et al.). For instance, both articles by Nicholas and colleagues (2007; 2008), generated similar themes despite focusing on different aspects of IBD. As limited reflexivity information was provided, it is impossible to determine the extent or nature of author contribution to theme development.

Strengths and Limitations

This is the first review focused exclusively upon CYP's experiences of IBD. This review only included studies that provided first-hand accounts of CYP experiences, and was strengthened by providing only direct participant quotes throughout synthesis findings. This ensured synthesis remained 'close' to the original studies, whilst also generating novel interpretations. Search sensitivity was strengthened by having no limitations on publications dates. Moreover, synthesis of qualitative studies provides nuanced and rich understandings of CYP's experiences of IBD, not capable of being achieved via quantitative enquiry, nor research amalgamating individuals across the lifespan. There was an almost equal spread of participants with UC and CD, with a broad age range and diverse disease severity, thereby increasing transferability of findings within the IBD population. The JBI tool allowed reviewers to appraise papers with greater consideration of interpretative and theoretical validity, in comparison to other tools (Hannes et al., 2010).

However, findings from this review must be considered within the context of its limitations. Inclusion of only English language and peer-reviewed studies does introduce publication bias. Studies included in this synthesis used diverse analytic approaches, which inevitably informed their analytic output. Although TS is capable of synthesising heterogeneous qualitative studies, it has been suggested that pooling findings in TS obscures the diversity of studies, which may represent a weakness in the current review (Lucas et al., 2007). Study quality was mixed, with some appraised as lower quality and lacking important methodological information. This influenced studies extent of contribution during synthesis, with those of higher methodological rigour and breadth of content inevitably contributing more so. For instance, quotes from both articles by Nicholas et al. (2007; 2008) featured quite prominently in synthesis write up; this was in part because they provided extensive quotations in their articles to illustrate findings, likely due to large sample sizes, but also as these studies were of relatively higher methodological quality.

Implications for Research & Clinical Practice

The variable quality of articles included in this review demonstrates scope for greater methodological rigour in qualitative research moving forwards. In particular, clear and explicit statements about researchers' epistemological position, philosophical perspective, and chosen methodology is necessary to ensure congruence; otherwise, qualitative reviewers are hindered in their ability to make informed judgements about the credibility of analytic outputs, a challenge experienced in undertaking this review. More transparent reporting of ethnicity is also required, alongside efforts to increase diversity of samples in terms of ethnicity and cultural contexts.

This review has highlighted the importance of language when communicating a ‘disease’ diagnosis to CYP, given the implications upon identity and emotional health. This terminology should be explicitly acknowledged at point of diagnosis, alongside HCPs exploring what this means to CYP on an individual basis, to ensure any misinterpretations can be resolved. Moreover, key adults are well-placed to foster CYP’s sense of agency in regards to living well with IBD and supporting CYP to retain their identity beyond this condition. HCPs and caregivers are encouraged to have open/anticipatory discussions with CYP about how they would prefer to be supported with IBD, differentiating support needs during periods of remission versus the event of a relapse.

The intruding sense of IBD upon CYP's BI is relevant to paediatric gastroenterology services, suggesting it may be beneficial for BI concerns to be routinely assessed to prevent more significant disturbances developing. Indeed, a recent SR found high prevalence of BI dissatisfaction in individuals with IBD across the lifespan (Beese et al., 2019). The IMPACT-III is a validated quality of life questionnaire developed for CYP with IBD that may assist with this as it includes a BI domain (Abdovic et al., 2013).

Conclusions

This review provided novel understandings of CYP's experiences of IBD, demonstrating nuanced differences from their adult counterparts. Findings have implications for those around CYP with IBD in terms of promoting adjustment to diagnosis and positive coping thereafter. It is imperative that support and interventions are tailored to the unique developmental implications of IBD for CYP if holistic, person-centered care is to be achieved. Study quality was diverse, demonstrating areas of methodological improvement required in qualitative research more broadly, to maximise credibility of findings.

References

Abdovic, S., Mocic Pavic, A., Milosevic, M., Persic, M., Senecic-Cala, I., & Kolacek, S. (2013). The IMPACT-III (HR) questionnaire: a valid measure of health-related quality of life in Croatian children with inflammatory bowel disease. *Journal of Crohn's and Colitis*, *7*(11), 908-915.

Barned, C., Fabricius, A., Stintzi, A., Mack, D. R., & O'Doherty, K. C. (2022). "The Rest of my Childhood was Lost": Canadian Children and Adolescents' Experiences Navigating Inflammatory Bowel Disease. *Qualitative Health Research*, *32*(1), 95-107.

Barned, C., Stinzi, A., Mack, D., & O'Doherty, K. C. (2016). To tell or not to tell: A qualitative interview study on disclosure decisions among children with inflammatory bowel disease. *Social Science & Medicine*, *162*, 115-123.

Barnes, C., Ashton, J. J., Borca, F., Cullen, M., Walker, D. M., & Beattie, R. M. (2020). Children and young people with inflammatory bowel disease attend less school than their healthy Peers. *Archives of Disease in Childhood, 105*(7), 671-676.

Barnes, E. L., Loftus Jr, E. V., & Kappelman, M. D. (2021). Effects of race and ethnicity on diagnosis and management of inflammatory bowel diseases. *Gastroenterology, 160*(3), 677-689.

Barnett-Page, E., & Thomas, J. (2009). Methods for the synthesis of qualitative research: a critical review. *BMC Medical Research Methodology, 9*(1), 1-11.

Beese, S. E., Harris, I. M., Dretzke, J., & Moore, D. (2019). BI dissatisfaction in patients with inflammatory bowel disease: a systematic review. *BMJ Open Gastroenterology, 6*(1), e000255.

Brooks, A. J., Rowse, G., Ryder, A., Peach, E. J., Corfe, B. M., & Lobo, A. J. (2016). Systematic review: psychological morbidity in young people with inflammatory bowel disease–risk factors and impacts. *Alimentary Pharmacology & Therapeutics, 44*(1), 3-15.

Camara, M., Bacigalupe, G., & Padilla, P. (2017). The role of social support in adolescents: are you helping me or stressing me out?. *International Journal of Adolescence and Youth*, *22*(2), 123-136.

Carroll, C. (2017). Qualitative evidence synthesis to improve implementation of clinical guidelines. *BMJ*, 356.

Cervesi, C., Battistutta, S., Martelossi, S., Ronfani, L., & Ventura, A. (2013). Health priorities in adolescents with inflammatory bowel disease: physicians' versus patients' perspectives. *Journal of Paediatric Gastroenterology and Nutrition, 57*(1), 39-42.

Clarke, T., & Lusher, J. (2016). Transitioning patients with inflammatory bowel disease (IBD) from adolescent to adult services: a systematic review. *Frontline Gastroenterology, 7*(4), 264-270.

Cosnes, J., Gower–Rousseau, C., Seksik, P., & Cortot, A. (2011). Epidemiology and natural history of inflammatory bowel diseases. *Gastroenterology, 140*(6), 1785-1794.

Erikson, E. (1968). *Identity: Youth in Crisis* (p. 94). New York: W.W. Norton.

Flemming, K., Booth, A., Hannes, K., Cargo, M., & Noyes, J. (2018). Cochrane Qualitative and Implementation Methods Group guidance series—paper 6:

reporting guidelines for qualitative, implementation, and process evaluation evidence syntheses. *Journal of Clinical Epidemiology, 97*, 79-85.

Fourie, S., Jackson, D., & Aveyard, H. (2018). Living with inflammatory bowel disease: a review of qualitative research studies. *International Journal of Nursing Studies, 87*, 149-156.

Griffiths, A. M. (2004). Specificities of inflammatory bowel disease in childhood. Best practice & research. *Clinical Gastroenterology, 18*(3), 509-523.

Hannes, K., Lockwood, C., & Pearson, A. (2010). A comparative analysis of three online appraisal instruments' ability to assess validity in qualitative research. *Qualitative Health Research, 20*(12), 1736-1743.

Holaday, B., Brydolf, M., & Segesten, K. (1996). "They feel your needs in the air": experiences of supportive activities among adolescents with ulcerative colitis. *Journal of Paediatric Nursing*, *11*(1), 71-78.

IBD UK. (2019). *Paediatrics and the IBD Standards.* Retrieved May 1, 2022 from www.ibduk.org/blog/2019/paediatrics-and-the-ibd-standards.

Keeton, R. L., Mikocka-Walus, A., & Andrews, J. M. (2015). Concerns and worries in people living with inflammatory bowel disease (IBD): A mixed methods study. *Journal of Psychosomatic Research*, *78*(6), 573-578.

Knowles, S. R., Graff, L. A., Wilding, H., Hewitt, C., Keefer, L., & Mikocka-Walus, A. (2018a). Quality of life in inflammatory bowel disease: a systematic review and meta-analyses—part I. *Inflammatory Bowel Diseases, 24*(4), 742-751.

Knowles, S. R., Keefer, L., Wilding, H., Hewitt, C., Graff, L. A., & Mikocka-Walus, A. (2018b). Quality of life in inflammatory bowel disease: a systematic review and meta-analyses—part II. *Inflammatory Bowel Diseases*, *24*(5), 966-976.

Kübler-Ross, E., & Kessler, D. (2005). *On grief and grieving: Finding the meaning of grief through the five stages of loss*. Simon and Schuster.

Lancet, T. (2012). Inflammatory bowel diseases audited. *Lancet (London, England), 379*(9819), 868.

Lindfred, H., Saalman, R., Nilsson, S., Sparud-Lundin, C., & Lepp, M. (2012). Self-reported health, self-management, and the impact of living with inflammatory bowel disease during adolescence. *Journal of Paediatric Nursing*, *27*(3), 256-264.

Lockwood, C., Munn, Z., & Porritt, K. (2015). Qualitative research synthesis: methodological guidance for systematic reviewers utilizing meta-aggregation. *JBI Evidence Implementation*, *13*(3), 179-187.

Lucas, P. J., Baird, J., Arai, L., Law, C., & Roberts, H. M. (2007). Worked examples of alternative methods for the synthesis of qualitative and quantitative research in systematic reviews. *BMC Medical Research Methodology*, *7*(1), 1-7.

MacKner, L. M., Crandall, W. V., & Szigethy, E. M. (2006). Psychosocial functioning in pediatric inflammatory bowel disease. *Inflammatory Bowel Diseases, 12*(3), 239-244.

McLoughlin, R. J., Klouda, A., Hirsh, M. P., Cleary, M. A., Lightdale, J. R., & Aidlen, J. T. (2020). Socioeconomic disparities in the comorbidities and surgical management of paediatric Crohn's disease. *Paediatric Research, 88*(6), 887-893.

Moorman, E. L., Koskela-Staples, N. C., & Janicke, D. M. (2023). A Systematic Review of Sleep Disturbances in Paediatric Inflammatory Bowel Disease. *Journal of Paediatric Psychology*, 48(3), 267-282., Chicago

Munn, Z., Stern, C., Aromataris, E., Lockwood, C., & Jordan, Z. (2018). What kind of systematic review should I conduct? A proposed typology and guidance for systematic reviewers in the medical and health sciences. *BMC Medical Research Methodology, 18*(1), 1-9.

Muse, K., Johnson, E., & David, A. L. (2021). A feeling of otherness: a qualitative research synthesis exploring the lived experiences of stigma in individuals with inflammatory bowel disease. *International Journal of Environmental Research and Public Health, 18*(15), 8038.

NHS. (2020). *Inflammatory Bowel Disease*. Retrieved July 8, 2022, from https://www.nhs.uk/conditions/inflammatory-bowel-disease/

Nicholas, D. B., Otley, A., Smith, C., Avolio, J., Munk, M., & Griffiths, A. M. (2007). Challenges and strategies of children and adolescents with inflammatory bowel disease: a qualitative examination. *Health and Quality of Life Outcomes, 5*(1), 1-8.

Nicholas, D. B., Swan, S. R., Gerstle, T. J., Allan, T., & Griffiths, A. M. (2008). Struggles, strengths, and strategies: an ethnographic study exploring the experiences of adolescents living with an ostomy. *Health and Quality of Life Outcomes*, *6*, 1-8.

Olsen, I. Ø., Jensen, S., Larsen, L., & Sørensen, E. E. (2016). Adolescents' lived experiences while hospitalized after surgery for ulcerative colitis. *Gastroenterology Nursing*, *39*(4), 287.

Plevinsky, J. M., Gumidyala, A. P., Poulopoulos, N., Kahn, S. A., Guilfoyle, S. M., Hommel, K. A., & Greenley, R. N. (2018). Illness-related parenting stress among parents of adolescents and young adults with inflammatory bowel diseases. *Children's Health Care*, *47*(2), 136-149.

Ponder, A., & Long, M. D. (2013). A clinical review of recent findings in the epidemiology of inflammatory bowel disease. *Clinical Epidemiology, 5*, 237.

Raza, A., Chan, V., & Cheema, M. A. (2019). Atypical Clinical Presentation of Crohn’s Disease as Diffuse Abdominal Lymphadenopathy and Venous Thrombosis: A Difficult to Diagnose Case. *Cureus*, *11*(9).

Reichenberg, K., Lindfred, H., & Saalman, R. (2007). Adolescents with inflammatory bowel disease feel ambivalent towards their parents’ concern for them. *Scandinavian Journal of Caring Sciences*, *21*(4), 476-481.

Shaw, R. L., Booth, A., Sutton, A. J., Miller, T., Smith, J. A., Young, B. & Dixon-Woods, M. (2004). Finding qualitative research: an evaluation of search strategies. *BMC Medical Research Methodology, 4*(1), 1-5.

Shorey, S., & Ng, E. D. (2020). The lived experiences of children and adolescents with non-communicable disease: A systematic review of qualitative studies. *Journal of Paediatric Nursing*, *51*, 75-84.

Sun, Y., Li, L., Xie, R., Wang, B., Jiang, K., & Cao, H. (2019). Stress triggers flare of inflammatory bowel disease in children and adults. *Frontiers in Paediatrics, 7*, 432.

Tempchin, J., Storch, B., & Reigada, L. C. (2021). Systematic review: Psychosocial factors of resilience in young people with inflammatory bowel disease. *Journal of Psychosomatic Research*, *148*, 110558.

Thomas, J., & Harden, A. (2008). Methods for the thematic synthesis of qualitative research in systematic reviews. *BMC Medical Research Methodology, 8*(1), 1-10.

Tran, L., & Mulligan, K. (2019). A systematic review of self-management interventions for children and adolescents with inflammatory bowel disease. *Inflammatory Bowel Diseases, 25*(4), 685-698.

Tong, A., Flemming, K., McInnes, E., Oliver, S., & Craig, J. (2012). Enhancing transparency in reporting the synthesis of qualitative research: ENTREQ. *BMC Medical Research Methodology, 12*(1), 1-8.

Vejzovic, V., Bramhagen, A. C., Idvall, E., & Wennick, A. (2018). Swedish children's lived experience of ulcerative colitis. *Gastroenterology Nursing*, *41*(4), 333-340.

World Health Organisation. (2016). *Adolescent Health.* Retrieved 1 February, 2023 from www.who.int/health-topics/adolescent-health#tab=tab_1

Woodgate, R. L., Tennent, P., Barriage, S., & Legras, N. (2022). The centrality of disclosure decisions to the illness experience for youth with chronic conditions: a qualitative study. *Journal of Health Psychology, 27*(3), 521-533.

Chapter 2: Major Research Project

Exploring Infant Mental Health in the Congenital Heart Disease Population: A Thematic Analysis

Prepared in accordance with the author requirements for:

Infant Mental Health Journal

www.onlinelibrary.wiley.com/page/journal/10970355/homepage/forauthors.html

Plain Language Summary

Title

Exploring the mental health of infants who have had surgery for Congenital Heart Disease (CHD)

Background

CHD is a very common birth defect in the UK and one third of infants with this condition will require surgery. Being hospitalised for surgery means the infant will be in an unfamiliar medical environment and experience frequent painful procedures, all whilst separated to some extent from their parents/caregivers. So far, there has been limited research about the potential impact of hospitalisation and surgery on the infants' social and emotional wellbeing, e.g. their mental health (MH), as research has instead focused on caregiver experiences or older children with CHD.

Aims and Questions

This study aims to explore:

1. What parents/caregivers think of their infant's MH after hospital and surgery for CHD,
2. Whether there were any aspects of hospital or surgery that parents/caregivers thought impacted the infants MH,
3. How parents/caregivers found completing a questionnaire about infant MH; in particular, how they found completing it themselves, the relevance of questions to infants with CHD, and their thoughts on it being used more widely in cardiac services.

Methods

Clinicians from the Cardiac Service identified potential participants and provided them information about the study. Seven mothers of infants with CHD, aged between 1-3 years old, took part in semi-structured interviews and completed a questionnaire about infant MH. Reflexive Thematic Analysis was used to develop themes from across the interviews. The questionnaires were scored to provide further information about infants' MH.

Main Findings

Parents/caregivers believed that infants who had had surgery in the first few weeks-months of life hadn't been aware of or affected by this. For infants that had had surgery later, parents/caregivers felt the infants' MH improved post-surgery, once settled home. However, they did notice some lasting effects from being hospitalised, including difficulties with feeding and sleep, changes in how the infant acted towards them and others, and fears about medical procedures, health professionals or clinical environments. Parents/caregivers identified various aspects of hospital that they considered to have either been helpful for the infants' MH or detrimental to it. They were accepting of completing the questionnaire, and preferred doing so themselves, but expressed if they had been given this by a medical professional their reaction may have been different. Some parents/caregivers assumed something must be ‘wrong’ with their infant if a medical professional were to give them such a questionnaire. They explained that how the questionnaire is introduced to parents/caregivers and when it is done are important considerations if it was to be used more widely.

Conclusions

This study deepens understandings about the MH of infants with CHD who have had surgery and a hospital stay. This study identified various aspects of hospital that could be improved to make these experiences easier for infants and their families. Although the questionnaire was generally accepted by parents/caregivers, and sensitive to the difficulties of infants with CHD, further research assessing the questionnaire more formally would be beneficial.

Abstract

Background: Congenital Heart Disease (CHD) is a common birth defect and one third of infants with CHD require surgical intervention. Hospitalisation for surgery involves physical separation from caregivers, an aversive environment, and frequent invasive procedures; yet there is limited research about the impact of cardiac experiences upon infants' mental health (IMH). Measurement challenges present a barrier to exploring IMH in the CHD population. The Brief Infant Toddler Social Emotional Assessment (BITSEA) is a validated questionnaire that has been recommended as offering promise in routine clinical practice; however, use of this questionnaire in the CHD population is required in the first instance. **Aims:** This exploratory study aimed to develop an understanding of the impact of illness, hospitalisation, and cardiac surgery on the infants developing mental health (MH), as perceived by primary caregivers. This study also explored caregivers' experiences of completing an IMH questionnaire (the BITSEA), including their views on administration method, relevance of items, and perspectives about this being used more routinely in cardiac services. **Methods:** Seven primary caregivers engaged in semi-structured interviews and completed the questionnaire self-report. Data was analysed using Reflexive Thematic Analysis, adopting a critical realist stance. Descriptive information from the questionnaire further characterised caregiver reported IMH. **Results:** Analysis generated the following themes: *"Just a Baby" - Very Young Infants are Immune to Early Experiences; Improved IMH following Cardiac Surgery;* and *Lasting Impacts of Cardiac Experiences,* with three subthemes reflecting different facets of IMH. Factors perceived to have impacted the infants' MH during hospitalisation generated two themes: *Promoters of IMH* and *Destabilisers of IMH.* Two themes and one sub-theme were developed regarding experiences of the questionnaire: *IMH Questionnaire*

Accepted, Contingent on Explanation and Timing, and Self-Report Preferred, Promotes Processing and Honesty. These findings expand theoretical understandings of IMH in the CHD population and have clinical implications.

Introduction

Infant mental health (IMH) has been broadly defined as “healthy social and emotional development in the very early years” (The Scottish Government, 2020, p. 2). There is consensus that IMH reflects a complex interplay of biopsychosocial factors (Sameroff, 2000), contingent upon the development of early relationships (attachment) with primary caregivers (Ainsworth & Bowlby, 1991). Attachment is critical to infant survival and supports regulation of arousal (Sroufe, 2005). The quality of attachment interactions affects the infant's rapidly developing brain and is argued to be central to mental health (MH) in infancy and beyond (Balbernie, 2013). Attachment between caregivers and critically ill infants may be disrupted due to physical separation and stressors related to hospitalisation/illness (Bright et al., 2013; Jordan et al., 2014).

Congenital Heart Disease (CHD) is amongst the most common of birth defects in the UK and over one third of infants with CHD require hospitalisation and surgical intervention (Healthcare Improvement Scotland, 2019). Hospitalised infants often experience frequent painful procedures and intrusive sensory stimulation (McMahon & Chang, 2020), which is undoubtedly stressful for primary caregivers as well. These experiences can impact upon caregiver MH and their ability to attune and modulate the stress of this environment for the infant (McWhorter et al., 2022; Rempel et al., 2013). Research has predominantly focussed on mothers’ attachment to infants with CHD, with fewer studies of father-infant attachment (Bright et al., 2013). A recent systematic review of parents and infants with CHD demonstrated highly heterogeneous findings in terms of parent-infant bonding and attachment style, reflecting the complex interplay of factors shaping this relationship (Tesson et al., 2022).

Research about the impact of CHD experiences upon infants is limited, with mothers having instead become the focus (Peters et al., 2019). Jordan and colleagues (2013) explored infant emotional wellbeing immediately after cardiac surgery, as perceived by mothers, and distinguished three groups: those perceived to be unaffected, those who were generally well but demonstrated specific fear responses to medical stimuli, and those with widespread emotional and/or behavioural dysregulation. Longitudinal research demonstrates high prevalence of emotional and behavioural difficulties in children who had had cardiac surgery in infancy, noting that these difficulties had been identifiable early on (Clancy et al., 2020). Routine assessment and monitoring of social, emotional, and behavioural development has been advocated for this population, to ensure early intervention and prevent longer term negative outcomes.

The routine assessment of IMH is impeded by measurement challenges. Observational strategies require training to administer, are labour-intensive and are therefore not suitable for routine clinical practice use. Moreover, there is debate around the ability to, and appropriateness of, classifying poor IMH as some difficulties may be contextual and/or transient (Szaniecki & Barnes, 2016). There is, however, a substantive body of longitudinal research that shows experiences in infancy, such as cardiac surgery, are predictive of later MH outcomes (Hövels-Gürich et al., 2007; Hughes et al., 2017). Specifically, children and young people (CYP) with CHD are at elevated risk of developing anxiety, depression, post-traumatic stress responses, and/or neurodevelopmental disorders (Gonzales et al., 2021), which further reinforces the need for time-efficient measures of IMH. Standardised tools may be advantageous for early identification and a recent review by Szaniecki and Barnes (2016) endorsed the Brief Infant-Toddler Social-Emotional Assessment (BITSEA; Briggs-Gowan & Carter, 2006) as a questionnaire of excellent psychometric quality with potential use in clinical practice. However, research exploring the use of such measures in the CHD population is required to evaluate suitability in this context.

Aims

Primary Aim: This study aimed to develop a deeper understanding about the impact of illness, hospitalisation, and cardiac surgery on the infants developing MH, as perceived by primary caregivers.

Secondary Aim: This study also aimed to gather primary caregivers' experiences of completing a questionnaire focussed on IMH.

Research Questions (RQs):

1. How do primary caregivers perceive the impact of hospitalisation and cardiac surgery upon their infant's MH?
2. What factors, if any, do primary caregivers perceive impacted their infant's emotions, behaviours or relationships during hospitalisation and surgery?
3. What are primary caregivers' experiences of completing a questionnaire focussed on their infant's emotions, behaviours, and relationships? Specifically, what are their views on the method of administration and the relevance of these questions in relation to the infant CHD population?

Methods

This study was reported in accordance with the Consolidated Criteria for Reporting Qualitative Studies (COREQ; Tong et al., 2007; see Appendix F, p. 125). The original project proposal can be found in Appendix G (p. 127).

Design

A qualitative-dominant, cross-sectional design was employed using semi-structured interviews to collect caregivers' perceptions of their infant's MH and their experiences of a questionnaire. Quantitative data from the questionnaire provided complementary descriptive information to further characterise the caregiver perceived IMH. Reflexive Thematic Analysis (RTA; Braun & Clarke, 2021a), adopting a critical realist stance, was considered appropriate as the present study sought to explore the primary caregivers' inferences, assumptions, and beliefs about their infant's MH, as opposed to being a direct reflection of the infants' experiences. As critical realism locates itself between positivist and constructionist paradigms, it allows for cautious interpretations about the infant's MH to be generated, whilst also acknowledging the context in which these are being constructed (i.e. the researcher's interpretations of the caregivers' perceptions, mediated by cultural context and individual characteristics).

Participants and Recruitment

Purposive sampling was used to recruit primary caregivers of infants who had been discharged home from the Glasgow Royal Hospital for Children (RHC) in the 18 months previous following cardiac surgery. An 18 month period since discharge was chosen to facilitate recruitment and feasibility, whilst also promoting participants ability to recall their infant's experiences. This timeframe minimised opportunities for other significant experiences to have occurred that could potentially have impacted upon IMH. Guidance from Braun and Clarke (2013) recommends between 6-10 participants for interview studies using RTA, as this permits important patterns of shared meaning across the dataset to be generated and a coherent narrative in relation to the RQ.

The inclusion of non-traditional families was permitted in aim of expanding the focus of IMH research beyond the mothers' perspective. Eligible participants were therefore any primary caregiver of an infant with CHD aged between 1-3 years old at the time of the interview, to allow for the completion of the BITSEA. Infants must have had at least one cardiac surgery and associated hospitalisation. Primary caregivers had to be able to speak sufficient English to participate in interviews. If more than one primary caregiver wished to participate, this was permitted. Infants who had comorbid diagnoses of neurodevelopmental disorders and/or genetic syndromes were excluded as these may have differentially impacted upon IMH. Infants whose physical condition was deemed at risk of sudden deterioration and/or poor prognosis were also excluded to avoid additional burden or stress for families. Extended family members without a primary caregiver role were not eligible.

Recruitment occurred in the Cardiology Department of Glasgow's RHC over a seven-month period. First, eligible caregivers were identified by members of the cardiac team. At a routine clinical interaction, clinicians provided caregivers with information about the study and the participant information sheet (PIS, see Appendix H, p. 128). The lead researcher then liaised with those who had consented to contact, to provide further information and arrange an interview, where appropriate. Contact occurred via email, telephone, or both, dependant on which communication format caregivers consented to. Participants were offered the option of in person or remote interviews and given a £10 shopping voucher to compensate for their time. Recruitment, transcription, and initial coding occurred concurrently, which aided decision-making of when sufficient data had been gathered to move from coding to the generation of meaningful themes, thereby ceasing recruitment (Braun & Clarke, 2019).

Thirteen caregivers consented to contact and nine of these agreed to take part; however, two were then unable to proceed due to other commitments. The four who did not take part did not respond to contact efforts and so it was not possible to ascertain reasons for not participating. It is likely that recruitment was negatively impacted by occurring over the festive period. The final sample included seven participants.

Materials & Measures

Quantitative

Demographic information about the primary caregiver and infant was collected. Infants' specific cardiac diagnoses and surgery type has not been reported on an individual basis to protect participant confidentiality.

The BITSEA is a 42-item questionnaire standardised for infants aged 12-36 months and aims to identify social-emotional and behavioural problems or delays in competence (Briggs-Gowan & Carter, 2006; Appendix I, p. 129). The BITSEA generates two total scores: a 'problem' total score and a 'competence' total score. Infants whose total 'problem' score falls into the 75th percentile or above are at greater risk of social-emotional difficulties. Infants whose total 'competence' scores falls into the 15th percentile or below are at greater risk of some form of deficit/developmental delay. The BITSEA has robust psychometric properties in terms of test-retest reliability, inter-rater reliability, and predictive validity (Briggs-Gowan et al., 2013; Kruizinga et al., 2011).

Qualitative

Semi-structured interviews gathered qualitative information about primary caregivers' perceptions of their infant's MH following hospitalisation for cardiac surgery, perceived factors that influenced this (if any), and their experiences of completing the BITSEA. An interview schedule (Appendix J, p. 130) was informed by relevant literature and developed in consultation with the Cardiology Service and a service user group (The Scottish Association for Children with Heart Disorders) to ensure questions were accessible, sensitive, and appropriate. A semi-structured format afforded flexibility in discussion, which was deemed imperative given the sensitive and emotive experiences being discussed. Reflective notes were made after each interview to aid reflexivity.

Research Procedures

All participants opted for remote interviews via Microsoft Teams, which facilitated childcare of infants. The lead researcher completed interviews independently. Time was spent initially building rapport and discussing the PIS. Once informed consent had been provided (see Appendix K, p. 131), participants were asked to complete the BITSEA via an online survey tool. Despite potential priming effects, it was anticipated that the BITSEA would orient participants to the focus of the interview and relevant IMH concepts. The semi-structured interview followed and was recorded using the 'record' function on Microsoft Teams. Demographic information was collected lastly. The entire process lasted between 60-90 minutes. Recordings were saved to the lead researcher’s secure NHS account and then transcribed verbatim. Any potentially identifying details were omitted from transcripts. Transcripts were not checked by participants for comments/corrections. Recordings were kept for the duration of the study in case of Sponsor audit.

Data Analysis

Qualitative data was analysed using RTA, conducted manually on Microsoft Word. RTA considers themes as analytic outputs actively created by the researcher through interaction with the data, the researcher’s assumptions and knowledge, and analytic work (Braun & Clarke, 2021b). This method was considered suitable over others, such as Interpretative Phenomenological Analysis (Larkin et al., 2021), as the focus was not exploring caregivers own experiences of their infant’s hospitalisation and surgery; rather, the study sought to develop understandings of IMH across the cohort of infants with CHD, via caregivers' perceptions. In accordance with a critical realist stance, a predominantly inductive orientation informed analysis, with codes and themes being driven by the content of the data for the most part. The researcher’s knowledge of child development and MH allowed for semantic and latent codes to be developed, as it was not anticipated that participants would have the theoretical understanding or range in terminology to depict manifestations of IMH.

As recommended by Braun and Clarke (2006), analysis moved from data familiarisation and initial coding to the development of themes and review. Coding was undertaken by the lead researcher, with one coded interview being reviewed by the academic supervisor to sense check and consider other possible interpretations, as opposed to seeking consensus. Coding was conducted on a combination of paper and electronic transcripts, as the use of different formats aided novel insights (see Appendix L for a coding excerpt, p. 132). Once all interviews had been coded, the codes were then clustered by RQ, alongside relevant data extracts, which allowed for the development of candidate themes. Transcripts were then re-read, and codes reviewed to check the validity of candidate themes. This repeated engagement with the data allowed for themes to be refined further in terms of title and content. Thematic maps were developed by hand throughout analysis stages. Theme development was undertaken predominantly by the lead researcher, in ongoing collaboration with academic and field supervisors, and practitioners in the field. In accordance with a reflexive approach, there was no predetermined fixed end point to analysis (i.e. "saturation"; Low, 2019), as there is always the potential for new interpretations. Thus, an informed interpretative judgement was required to determine when the purpose and goals of the study had been sufficiently achieved (Braun & Clarke, 2021c).

Demographic information was collated. To prevent potential influence upon the qualitative analysis, the BITSEA questionnaires were intentionally scored after theme development. Raw scores were converted to standardized scores based on infants' gender and age. The frequency of infants within the sample scoring in the 'possible social-emotional problem' and 'possible deficit/developmental delay' range was reported alongside demographic information.

Reflexivity

RTA considers researcher subjectivity as a resource in qualitative research as opposed to a threat to be controlled (Braun & Clarke, 2021b). The lead researcher was a female Trainee Clinical Psychologist, meaning they were experienced in conducting semi-structured interviews with individuals and/or family members experiencing emotional, social, and/or cognitive difficulties in MH services across the lifespan. Field and academic supervisors had doctoral level training in Clinical Psychology. The lead researcher had some existing experience of qualitative methods from a previous research role. The lead researcher undertook a placement at the RHC over the recruitment period of this study. This involved working with CYP whose MH had been affected by their physical health condition; however, they did not work directly with the Cardiology Service nor have any clinical experiences of CYP with CHD during this, thereby affording an ‘outsider’ perspective. It was important for the researcher to be mindful of their training in psychopathology, and the potential to seek out instances of MH difficulties in their questions asked and in reading of the data. Supervision and a reflective log helped to consider how the researcher’s values, experiences and beliefs influenced the meaning making process.

Ethics

Ethical approval was granted from the Southwest - Central Bristol NHS Research Ethics Committee (REC; IRAS312275) and managerial approval from NHS Greater Glasgow and Clyde’s Research and Innovation Department (GN22MH191P; see Appendices M & N, p. 133-139).

Results

Full details of participant demographic information are provided in Appendix O (p. 140). All seven primary caregivers who participated were mothers of infants with a range of CHD diagnoses, such as septal defects, aortic valve stenosis, patent ductus arteriosus and others. Severity of CHD differed amongst the sample, reflected in the variable total time spent in hospital and number of surgeries to date. At the time of interviews, infants had a mean age of 24 months (youngest 16 months; oldest 31 months) and had been discharged home from hospital a minimum of 8 weeks prior and a maximum of 1 year ago. 42.9% (*n* = 3) of the infants scored in the 'possible social-emotional problem' range within the BITSEA and 14.3% (*n* = 1) scored in the ‘possible deficit/developmental delay’ range.

Reflexive Thematic Analysis

Table 1 provides an overview of the analytic outputs of RTA, according to each RQ. Themes and subthemes are discussed further below, alongside illustrative quotes. Table 2 details the age of each infant (and assigned caregiver pseudonym) to help contextualise the findings according to infants' developmental stage.

Table 1

Overview of Themes and Subthemes

Research Question	Themes and Subthemes	Brief Description
RQ1: How do primary caregivers perceive the impact of hospitalisation and cardiac surgery on their infant's MH?	1. *"Just a Baby"* - Very Young Infants are Immune to Early Experiences	Caregivers perceived that their infants had been entirely unaffected by and had no memory of early cardiac experiences, implying an assumed age at which IMH develops.
	2. *"A Different Baby"* - Improved IMH following Cardiac Surgery	A significant improvement in infant's overall wellbeing & MH following cardiac surgery and discharge home was perceived. This prompted caregiver comparisons to how the infant's MH had been prior to surgery.
	3. Lasting Impacts of Cardiac Experiences Feeding Difficulties and Sleep Issues Changes in Infants Relational Style Medical Fear Responses	Despite reporting a general improvement in IMH, caregivers also identified specific aspects of the infant's MH that they perceived had been affected by cardiac experiences, delineated by three sub-themes.
	4. Promoters of IMH	

RQ2: What factors, if any, do primary caregivers perceive impacted their infant’s emotions, behaviours or relationships during hospitalisation and surgery?	5. Destabilisers of IMH	Caregivers identified various experiences and aspects of the infant’s hospitalisation that they felt had either promoted or threatened the infant’s developing MH.
RQ3: What are primary caregivers’ experiences of completing a questionnaire focussed on their infant’s emotions, behaviours, and relationships?	6. IMH Questionnaire Accepted, Contingent on Explanation & Timing Psychopathology Assumed from a Questionnaire	Caregivers were accepting of the questionnaire; however, caveated that this would be contingent upon timing of completing this, and explanation/rationale provided. Some caregivers assumed that being given a questionnaire meant psychopathology must be suspected.
	7. Self-Report Preferred, Promotes Processing and Honesty	Caregivers preferred completing the questionnaire independently. This format afforded greater consideration of their responses and promoted a more honest response style.

Table 2

Contextual Information regarding Infants

Caregiver Pseudonym (Interview)	Infant Age
Beth (1)	31 months
Rachel (2)	17 months
Louise (3)	24 months
Sara (4)	35 months
Emma (5)	20 months
Fiona (6)	24 months
Kate (7)	16 months

Theme 1: *"Just a Baby" - Very Young Infants are Immune to Early Experiences*

Some infants included in this sample had had lengthy hospitalisations and cardiac surgery in the first few weeks-months of life and then again at a later stage of infancy. Reflections on infants earlier cardiac experiences were notably absent from caregivers' accounts or only briefly mentioned, due to beliefs that these experiences did not have any relevance to IMH nor lasting impacts. This was evident across participants, for example*: "He was too young, so he won't remember*" (Louise), and *"The first-time round, he was only a baby, so I don't think it really had, overly, much of an effect on him"* (Rachel). These descriptions suggest an underlying assumption that very young infants are initially immune to their experiences. This was communicated in relation to different facets of IMH, including beliefs that infants at this age have no awareness nor sensitivity to their external world, are unable to learn or develop memories, and don't have the capacity for emotional development. This implies an assumed age within infancy at which IMH begins to develop and only then is affected by cardiac experiences. Consider this extract from Kate:

> *"The first time [surgery] didn't phase her because, just being so small, it was just you know, at that age of all the cat napping and feeding"*

This denotes very young infants as having only physiological needs to be met (i.e. eat, sleep, repeat). Descriptions such as *"He was just a baby"* unintentionally negate that infants are sentient and have a 'MH' that is shaped by early experiences, interactions and their environment. It was not within the realm of the interviews to explore where such ideas stemmed from; however, one possibility is that these beliefs afforded a protective psychological defence, given how distressing and difficult it may be for caregivers to consider the potential psychological impacts of critical medical experiences. Alternatively, it may reflect understandings of IMH held more broadly by society.

Theme 2: *"A Different Baby" - Improved IMH following Cardiac Surgery*

Caregivers reported an overall improvement in their infant's MH following surgery, once infants were beyond the immediate recovery phase and discharged home. This was particularly apparent in participants whose infants had had their first cardiac surgeries slightly later in infancy, as opposed to within the first few weeks-months of life. This may be because those caregivers had not known the infant to present in any other way and so were more likely to report perceptions resembling Theme 1. Observations of how the infant behaved and interacted with the world following cardiac surgery prompted caregivers to reflect on the infant's MH prior to this. Caregivers described their infants pre-surgery as having a greater need for proximity, being unable to soothe or rejecting their efforts, and displaying less enjoyment from activities compared to their peers; whereas, post-surgery, infants were perceived to express more positive affect, have increased confidence and exploration, and were generally more responsive and engaged in life.

> *"He just wanted cuddled, breast fed, carried everywhere. Umm… he was just obviously miserable [pre-surgery]. But now [post-surgery] he's just like, 'No, [infant] do it. [Infant], do it. No, mummy'. Umm, so yeah, he's much more excited*" (Beth)

Post-surgery, caregivers welcomed age-appropriate social-emotional behaviours with discernible triggers, for example: "*I think she's much more content and yeah it's nice to see like the normal tantrums rather than the things we've seen before, where she was like upset for no real reason or just like in general*" (Sara). Clear distinctions were made between the infant's MH and identity before and after surgery, as illustrated in this extract from Louise:

> *"Me and my husband say that when he got that surgery and we came home, we had a different baby. So it's like that surgery date we see as like a […] birthday as well for us because he was like a different child or baby after he got his heart fixed"*

Participants depicted a stalling of the infants' developmental milestones during the acute illness phase, hospitalisation, and surgery; whereas once infants were home after surgery, development reportedly rapidly advanced. For example, Fiona described:

> *"Once it got fixed [heart] and she came home, there was no stopping her. She just had so much more energy, you could just see the difference in her. And she probably did become more animated, she was*

> *happy before, but she was a lot more happier after her surgery. Wasn't as tired, and her development just went through the roof. She just liked walked, starting to try and move more, speech got better, yeah, she was trying to do loads"*

Accounts conveyed instantaneous transformations in the infants' presentation, which surprised caregivers and marked a new beginning for the infant and family. Implicit within caregivers' descriptions was some understanding of the mind-body connection, as IMH improvements were, in part, attributed to physical health improvements.

Theme 3: *Lasting Impacts of Cardiac Experiences*

Despite the general, overall improvement in IMH following surgery, caregivers' also highlighted specific aspects of the infant's MH that they perceived had been affected by illness, hospitalisation, and/or surgery. The following three sub-themes represent three aspects of IMH that caregivers referred to.

Subtheme 1: *Feeding Difficulties and Sleep Issues*

Caregivers described lasting impacts from hospitalisation/surgery on the infants' feeding practices and sleeping routines. Many of the infants had experienced NG-tube feeding during hospitalisation and caregivers frequently referenced difficult experiences with this as contributing to oral aversions thereafter. Rachel described "*I think the feeding thing just had such a major influence on him. And as I say, I think it's still affects him even now. Em… I think that's probably been the most major thing"*. Caregivers related the success of infants' feeding to their MH, perhaps alluding to the fact that feeding goes beyond providing nourishment to the infant and is also a means of offering containment and soothing that infants were then unable to access and benefit from. Others described lasting impacts of hospitalisation/surgery upon their infants' sleep: *"She had started getting like really, really, troubled sleeping after that [surgery]. She's never been a great sleeper […] but we noticed a difference after that massively"* (Emma).

Subtheme 2: *Changes in Infants' Relational Style*

Participants reported differences in how their infant related to them and others following hospitalisation/surgery. Some caregivers described the emergence of separation anxieties, whereby the infant would become inconsolable during separations and appear mistrusting of others. For example:

> *"He never used to be that clingy to us at all. He would just go with anybody, he was no bother. Even in the house, he's like my wee shadow. He just follows me around everywhere and he'll go away for like 2 minutes and then he's back... I don't know, I don't know if that's anything to do with everything he's had, I don't know, it might be"* (Rachel)
>
> *"If I have to leave the house he will scream, even for my husband. Only a couple of months ago he screamed for like an hour straight because I left him"* (Louise)

These infants were increasingly reliant on their caregivers and sought out greater proximity following hospitalisation. The need for proximity also permeated sleep routines and portrayed possible fears of abandonment following hospitalisation:

> *"When she's falling asleep, she does like to lie on you, which she never done before [surgery]. She always, she didn't like cuddling in to sleep previously but now she likes to lay on top of me knowing that I can't move and get away from her kind of thing" (Emma).*

Conversely, two participants described their infants as highly sociable and interaction-seeking following hospitalisation for surgery, which they attributed to the extent of interactions and tactile experiences they had had with numerous health care professionals (HCPs) during hospitalisation. Of note, both infants whose caregivers reported this relational style had had surgery in the first few weeks of life as opposed to later in infancy, which may contribute to the differential impacts.

Subtheme 3: *Medical Fear Responses*

Throughout the analytic process, the medical fear responses that infants developed following cardiac experiences appeared particularly prominent. These responses involved aversion, avoidance, hypervigilance and overt distress, akin to modern understandings of paediatric medical traumatic stress (PMTS; National Child Traumatic Stress Network, 2003). Caregivers described how infants communicated their aversions to HCPs and certain medical equipment via their behaviour and, or verbally at times. This led to infants consistently trying to avoid, escape, or refuse to engage with HCPs and, or medical interventions.

> *"[Infant] is petrified of the nurses, still is scared of it. […] He's petrified of a uniform"* (Louise)
>
> *"'What are you going to do?!' it's like she knows that they're gonna do something and she's aware of what it is. I feel like she's just very wise and she's just waiting for something"* (Kate)
>
> *"Maybe because her experiences of hospital etc, she very much realises, as soon as she sees someone with a mask on (because of COVID), she very much turns into me and she knows that they are going to examine her […] We went to get her feet checked at the shoe shop*

> *and again she just cried, cause she knew this person was going to touch her"* (Fiona)
>
> *"When we go into the hospital, he knows where we're going and as soon as we go into the room, he starts getting upset usually. Em… because, he's just had so much to happen to him"* (Rachel)

Infants had developed negative associations with, and expectations of, unfamiliar adults in uniform and were vigilant to what HCPs were going to do, demonstrating mistrust and anxiety in their presence. Infants seemed particularly aversive to touch. Caregivers' descriptions conveyed a strong sense of agency from infants regarding their views and feelings about HCPs following hospitalisation/surgery:

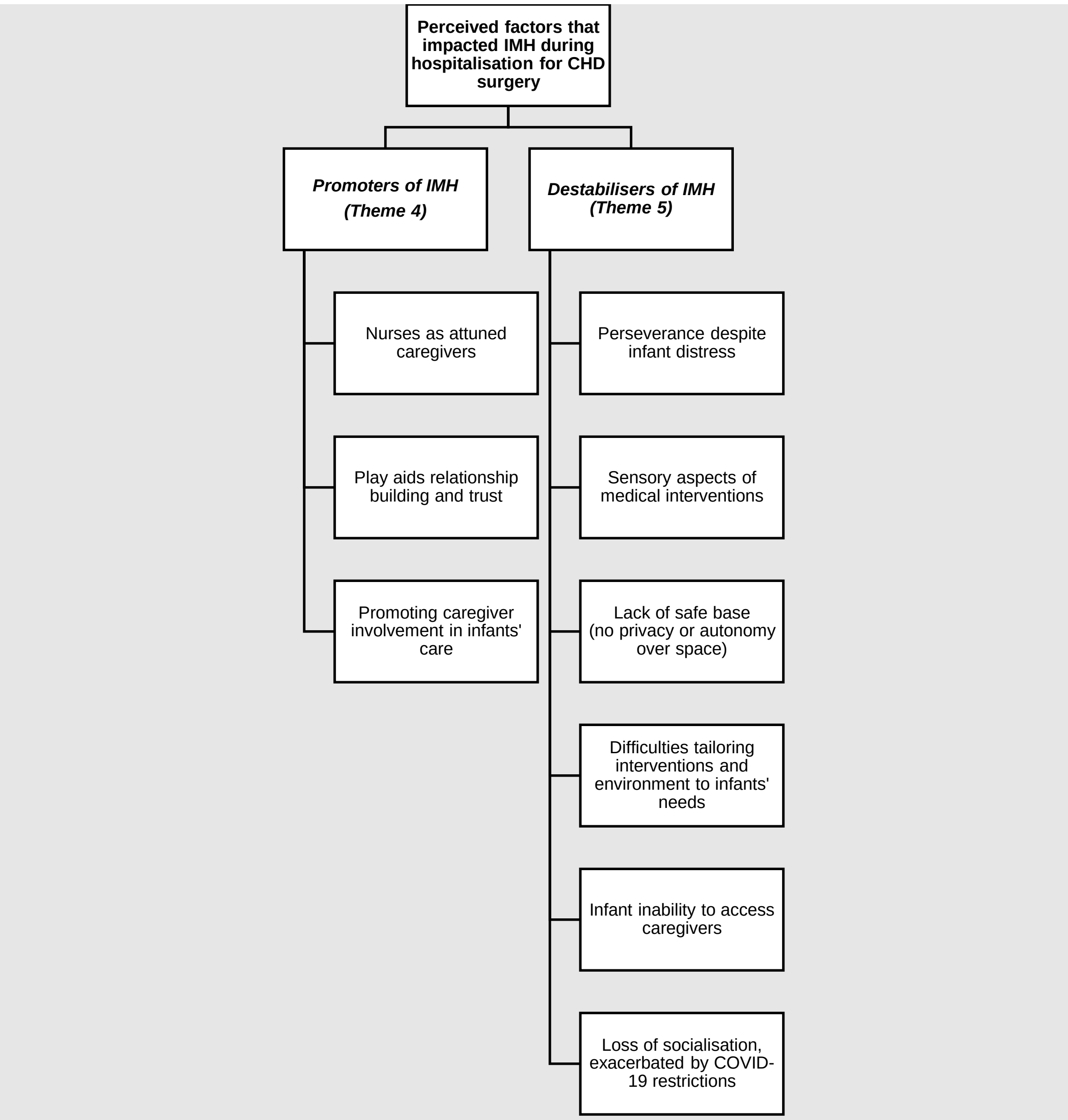
> *Interviewer: "How does he cope when he has to go and see the doctors then?"*
>
> *Beth: ""Go away. Go away Doc!!!""*

Themes 4 & 5: *Promotors and Destabilisers of IMH*

Certain aspects of the infants' hospital experience were particularly salient and relevant to their developing MH. These factors were understood as either promotors of IMH or destabilisers to the infants' MH. It should be noted that even factors caregivers associated with negative impacts (i.e. destabilisers) were caveated with gratitude for the medical care their infant had received. The specific factors identified within these two themes are summarised in Figure 1, and broadly related to environmental influences or qualities of interactions with HCPs. As these themes were consistent with existing findings in the CHD literature they are detailed further in Appendix P (p. 142).

Figure 1

Promoters and Destabilisers of IMH during Hospitalisation



Theme 6: *IMH Questionnaire Accepted, Contingent on Explanation & Timing*

Participants were accepting of the BITSEA and felt items were, for the most part, relevant to infants with CHD. Caregivers caveated however that the why, when, where and by whom were key considerations if this questionnaire were to be used more routinely with hospitalised infants. Caregivers emphasised the sensitive nature of timing, with certain time points in the infants' illness experience deemed inappropriate to complete this due to high emotional distress and changes in infant presentation at these times. Specifically, caregivers deemed it would not be appropriate to complete this during hospitalisation for cardiac surgery, rather, it would be helpful if used whilst an outpatient, either during a pre-operative appointment or once discharged home. Caregivers also highlighted that the rationale for the questionnaire would affect their reaction to this. Some caregivers viewed the questionnaire as potentially beneficial in facilitating early detection of IMH and intervention.

Subtheme 4: *Psychopathology Assumed from a Questionnaire*

Although the BITSEA was accepted for the purposes of research, some participants communicated assumptions that if HCPs had provided them with such a questionnaire whilst in hospital, then they must suspect there is something 'wrong' with the infant's MH. Assumptions that questionnaires are only used when there are indications of psychopathology evoked feeling evaluated or targeted as a parent. As Beth summarised: *"I think if it was one of the like junior doctors or nurses that came and did it, it might feel a bit like ohh right. What is wrong? [...] why does my child need assessed like mentally? Are you coming to judge my parenting?".* Caregivers were concerned about the implications or potential outcomes of such a questionnaire and indicated this could affect their response style if not clarified, as Emma described*: "I can imagine some people being like, 'Aw, what's going to happen if I give the wrong answers here?'".* This subtheme furthers understanding behind the caveats referenced in Theme 6 and has commonalities with Theme 1, suggesting an avoidance of thinking about IMH when physical health needs are in jeopardy (Lumsden et al., 2019).

Theme 7: *Self-Report Preferred, Promotes Processing and Honesty*

Self-report was consistently preferred by participants in terms of potential administration methods of the BITSEA. Participants reported that completing the questionnaire independently allowed them greater opportunity to reflect on their infant and consider their responses. This was important given participants described not often having opportunities to pause and reflect on their infant's behaviour or development to date, due to the busy demands of caregiving and additional CHD stressors. Emma also highlighted that self-report promotes a more honest response style, with others echoing that they would have been less forthcoming in their responses if the questions had been verbally asked to them; *"I think maybe filling it out yourself you are maybe more honest, instead of like a HCP".*

Discussion

This study explored IMH following hospitalisation and surgery for CHD via in-depth semi-structured interviews with primary caregivers. A secondary aim was to explore experiences of completing an IMH questionnaire. Three themes and three subthemes relating to the main RQ were developed using RTA:

1. *"Just a baby"* - Caregiver beliefs that very young infants and their developing MH are immune to early cardiac experiences
2. *"A different baby"* - Perceived improvements in IMH overall following later cardiac surgeries
3. *Lasting impacts of cardiac experiences* - Specific aspects of IMH were perceived to have been impacted by illness, hospitalisation, and/or surgery, including:
    - Feeding and sleeping difficulties
    - Changes in infants' relational style
    - Development of medical fear responses

In examining the second RQ, which focused explicitly on perceived emotional and behavioural impacts during hospitalisation for surgery, the two themes reflected the promoters and destabilisers relevant to the infant's developing MH. Finally, two themes, and one subtheme, were developed in relation to caregivers experience of completing the questionnaire:

1. Acceptance of the questionnaire, contingent on explanation & timing, as some caregivers reported:
    - Assumptions of psychopathology from a questionnaire
2. Preference for self-report.

"Just a Baby" - Very Young Infants are Immune to Early Experiences

Theme one demonstrated implicit beliefs caregivers held in relation to their infant's MH, which likely contributed to their perceptions, inferences, and reports of infants' cardiac experiences more broadly. Some infants were perceived as entirely unaffected, socially, emotionally, and perceptually, by very early cardiac experiences. This theme mirrors findings by Jordan et al. (2013) who found a proportion of very young infants with CHD were considered by caregivers to be "unscathed" by early cardiac experiences. This may indicate caregivers extent of mind-mindedness during this period, e.g. "caregivers' proclivity to treat their young children as individuals with minds of their own" (Meins et al., 2013, p. 1778); suggesting that caregivers of very young infants with CHD are less able to adopt a mind-minded stance during early hospitalisation and surgery for several reasons. Firstly, it is easier to recognise and understand an infant's mental states when the child's physical needs are met (Ellis, 2017), which is less possible whilst the infant is hospitalised and critically unwell, and at later stages of infancy when infants' social communication repertoire is more developed (Silletti et al., 2022). Although not specific to CHD, caregivers of pre-term infants experiencing high levels of stress demonstrated impaired mind-mindedness (Suttora et al., 2017). Such findings are relevant given infant CHD surgery constitutes a significant stressor for caregivers, which may, in turn, reduce their mentalisation capacity. Secondly, this could be understood within the hierarchy of needs framework (Maslow et al., 1970), whereby caregivers are unable to consider anything beyond the infant's immediate physical safety needs being met (i.e. their survival) during early cardiac experiences. Thus, this belief may serve as a necessary, psychological defence mechanism for caregivers to avoid emotional overwhelm during hospitalisation/surgery, a time known to be highly distressing (Woolf-King et al., 2017). Lastly, *"Just a baby"* conveys a detachment to the infant, which would make it more difficult to recognise their internal states; indeed, detachment has been found to be a coping strategy of parents of infants undergoing CHD surgery (Lumsden et al., 2019).

This theme also has implications in terms of understanding perceptions of IMH more broadly, suggesting there are disparities between the general population's perceptions (i.e. "*Just a Baby")* compared to contemporary understandings of IMH (Fitzgerald et al., 2011). Indeed, a systematic review, highlighted key differences in IMH understandings between researchers and parents of infants (Peters et al., 2019). Although the beliefs about IMH in this study came from a small homogenous sample of mothers, a recent study similarly found individuals were surprised to learn that infants have emerging capacities from birth and some had never previously considered the idea of 'IMH' (de Burca et al., 2022). Increasing caregiver understanding of MH in hospitalised infants with CHD is essential to promote the adaptation of social-environmental factors to better meet the infant's needs during this critical time of development.

"A Different Baby" - Improved IMH Overall following Cardiac Surgery, with some Specific Lasting Impacts

Themes 2 and 3 were also consistent with findings from Jordan et al. (2013, p. 262), whereby most infants with CHD were perceived by caregivers as "generally happy" following hospitalisation and surgery; however, with specific aspects of IMH then reportedly impacted by cardiac experiences. Taken together, these themes demonstrate the complex and nuanced impacts of cardiac experiences upon IMH that likely wouldn't have been understood via quantitative enquiry only. Indeed, infants' scores varied on the BITSEA, with an almost equal spread of infants who did or did not score in the 'possible social-emotional problem' range (42.9% & 57.1% respectively).

Parents perceived that IMH had improved overall once infants were discharged home following cardiac surgery, based upon increased expression of positive affect, demonstration of more 'typical' social-emotional behaviours, and rapid progression of developmental maturation. This helps to contextualise findings from the BITSEA, whereby fewer than 15% of infants scored in the 'possible deficit/developmental delay' range, so for this sample most developmental delays reportedly resolved post-surgery. It is plausible that IMH improved for various reasons once discharged home, including the infants' improved physical health, being cared for in a non-clinical, nurturing environment, and improved caregiver wellbeing and confidence (Gaskin, 2018).

Like Jordan et al.'s (2013) findings, caregivers in this study reported post-hospitalisation/surgery impacts such as feeding and sleep difficulties, changes in social and interpersonal behaviour, and negative emotional reactions in response to medical stimuli, including the presence of HCPs. Given the current sample included older infants, some of whom had had surgery up to one year prior, this suggests such difficulties are not transient. Medical fear responses have been identified in older children with CHD via qualitative (Chong et al., 2018) and quantitative (Gupta et al., 1998) enquiries. The various responses described following hospitalisation and surgery could be understood within the Integrative Model of Paediatric Medical Traumatic Stress (PMTS; Kazak et al., 2006). Although there is little research exploring PMTS in infants with CHD, CYP with CHD are at elevated risk of PMTS, with 12% of CYP meeting diagnostic criteria for PTSD 4-8 weeks after being discharged home from cardiac surgery and 31% of CYP continuing to meet criteria many years on (Meentken et al., 2017).

Within the current sample, there appeared to be two dominant infant relational styles following hospitalisation and surgery; some were perceived as more insecure in their relationships with caregivers and others, evidenced by greater need for proximity, maternal dependence and anxiety about interacting with others, in comparison to other infants that were highly independent and overly confident in their exploration. These findings may explain the high heterogeneity in attachment security that has been found between caregivers and infants with CHD (Tesson et al., 2022). Whilst the literature has predominantly focused on caregiver-related characteristics that may impact the developing attachment relationship, the current study provides novel insights to infants’ fluctuating interpersonal behaviour that has an active role in shaping the attachment relationship. Longitudinal research would be required to determine whether changes in the infants’ relational style are an adaptive, temporary response following hospitalisation, which the attachment-caregiving system can be resilient to, or whether these changes alter the infant-caregiver attachment style more permanently thereafter.

Promoters and Destabilisers of IMH during Hospitalisation and Surgery

Themes four and five represent the aspects of hospitalisation that caregivers perceived either promoted their infant’s MH or were considered detrimental to it. The ‘Promoters of IMH’ theme aligns with findings of Wei et al. (2017) regarding HCPs facilitative interactions, and the ‘Destabilisers of IMH’ resembled the ‘physical’ and ‘social’ risks identified by Ryan et al. (2019). The negative accumulative effect of such hospital-related factors upon infants’ neurodevelopment have been highlighted by Ryan and colleagues previously (2019), which have clinical implications that are discussed further below.

Experiences of the BITSEA

The infants’ scores on the BITSEA suggest that this IMH measure has sensitivity to potential difficulties within infants with CHD, although this was a preliminary evaluation with small sample size. Whilst caregivers were accepting of an IMH questionnaire for the most part, several important caveats to its potential future use in clinical care were identified. This echoes findings from other clinical health populations, whereby the potential benefits of assessment tools depend on the context of their use (Biddle et al., 2016). If used appropriately, caregivers recognised the potential benefits of such a measure for early detection and intervention for IMH in the CHD population. The measure itself may also help to raise awareness of IMH amongst staff and caregivers, thereby confronting beliefs outlined in Theme 1.

Strengths & Limitations

This study enacted recommendations by previous researchers in the field (Jordan et al., 2013; Dahlawi et al., 2020) by utilising in-depth qualitative enquiry, alongside other sources of evidence (e.g. the BITSEA), to provide a rich, multifaceted understanding of IMH in the cardiac population. The findings enhance theoretical understandings, with research to date having previously neglected the infants' cardiac experience by focusing on older children with CHD (Chong et al., 2018) or caregiver experience (Harvey et al., 2013). This study also highlighted clear implications for clinical practice, which, if acted upon, could mitigate the negative impact of certain cardiac experiences and promote IMH-informed care. Moreover, to our knowledge, this study is the first to trial the use a validated measure of IMH in the cardiac population and gather insights about the experiences of completing this. Our findings enable understanding of the factors determining the effective use of the BITSEA in clinical care; however formal acceptability/feasibility investigations are required in the CHD population.

Several limitations must be considered when interpreting the results of this study. Given the qualitative, cross-sectional nature of this project, it is impossible to determine whether some of the infants' difficulties would have developed regardless of cardiac experiences; however, in this sample, the caregivers clearly reported perceived impacts related to the infants' medical experiences. Despite ambitions to recruit a range of primary caregivers, as opposed to focusing on mothers only, findings were derived from a relatively small, homogenous sample of white mothers living in Scotland. Thus, caution should be taken in terms of the transferability of these findings to other contexts. A need remains to involve fathers or other primary caregivers more so in IMH research.

The range of cardiac diagnoses included within this sample increases transferability of findings within the CHD population; however, infants within this sample had varied cardiac experiences in terms of hospitalisation length and number of surgeries. It was not possible, nor the purpose of this exploratory study, to discern any potential differences in IMH according to diagnoses/nature of cardiac experiences.

Future Research Directions

Caregivers' MH, extent of knowledge/understanding about IMH, and their own experiences of hospitalisation were inextricably connected to and woven within their descriptions of the infants' experiences. The lead researcher's training in child and adult MH allowed them to remain cognisant of this, alongside a reflective log and regular supervision to maintain focus upon the infant's MH. This does however present an opportunity for future researchers to conduct secondary data analysis relating to the mothers' experiences included in this study. To the author's knowledge, there are no studies exploring the prevalence of medical fear responses, akin to PMTS, specifically in infants with CHD following surgery, which presents an opportunity for future research. Although there are studies exploring attachment style and the MH of older children with CHD (Gonzalez et al., 2021), it may be beneficial for future researchers to conduct longitudinal explorations of individuals with CHD from infancy into childhood and adolescence to determine the trajectory of the difficulties found in this study (i.e. changes in feeding/sleeping practices, relational style and development of medical fear responses). Lastly, this study offered preliminary evidence that the BITSEA was accepted by caregivers of infants with CHD and sensitive to their difficulties, however more formal feasibility studies would be advantageous prior to this being used more commonly in clinical practice.

Clinical Implications

Ryan et al. (2019) provide recommendations for clinical practice that are supported and extended further by the present study's findings. For instance, promoting caregiver involvement in the infants' care during hospitalisation by having caregivers present and in physical contact with the infant for any procedures, and/or assisting with administration of these where possible (e.g. caregiver holds stethoscope to infant's chest whilst HCPs listen). The environment and sensory input should be altered to be more nurturing and less clinical (e.g. monitoring and minimising noise levels on ward, dimming any bright lighting or having blackout spaces, access to white noise or children's music). Sensory aspects of medical interventions should also be adapted where possible (e.g. ensuring the gel used within echocardiograms is not extremely cold, if possible heat this in some way prior to applying to infants' skin, and/or ensuring the infant is distracted whilst blood pressure is being taken). Infants and families benefit from having access to a separate play space away from hospital room, promoting opportunities to meet and socialise with other infants and families. Separation from caregivers should be minimised by allowing caregivers to remain with their infant overnight pre and post-surgery. This is imperative to ensure the infant can access their caregiver for soothing during moments of distress. Care interactions should be clustered to minimise sleep disruption. One further practical suggestion would be to have a sign on the infant's hospital room door to alert HCPs whether they are sleeping and so shouldn’t be disturbed, if possible, or to enter as quietly as possible as this supports with quality sleep, dysregulation, and enables caregivers to establish less clinical routines that could ease the transition home post-surgery. Pain management should be carefully considered, to ensure a balanced approach that is infant-tailored and considers impact of pharmaceutical use on mental health. Where safe to do so, do not persevere with medical procedures if infants are extremely distressed and dysregulated. If HCPs are aware intervention or a procedure is going to be distressing or painful for the infant, ensure appropriate pain relief has been considered/administered and that they always have physical access to their caregiver during the procedure. Whilst the many clinical demands of HCPs are acknowledged, infants would benefit from even brief instances of playful interactions with HCPs throughout hospitalisation, as this enables trust building and encourages infants' co-operation and compliance with medical interventions thereafter. Access to bottle making facilities and a highchair available for mealtimes would support transition home upon discharge and encourage feeding rituals being established.

Conclusions

This study explored IMH following hospitalisation and surgery for CHD, via caregiver perception and a questionnaire, in aim of illuminating the infants' CHD experiences, which has been neglected to date. The findings provided important insight to caregiver beliefs about IMH more generally in the early months of infancy, a period whereby many infants with CHD will experience significant challenges such as extended hospitalisations and cardiac surgery. The findings also highlight the complex and nuanced impacts of cardiac experiences upon IMH, as perceived by caregivers, which have implications for the developing infant-caregiver attachment relationship. Consistent across infants within this study was the development of medical-fear responses, akin to PMTS, which has implications for clinical care and should be a focus of future research. Various aspects of the infants' medical care were identified as either facilitative or detrimental to IMH, highlighting potential areas for service development. Lastly, the BITSEA questionnaire demonstrated potential for use in clinical practice to ensure early detection and intervention for IMH difficulties, such as those found in this study; however, further acceptability/feasibility research would be advantageous.

References

Ainsworth, M. D. S., & Bowlby, J. (1991). An ethological approach to personality development. *American Psychologist, 46*, 331-341.

Balbernie, R. (2013). The importance of secure attachment for IMH. *Journal of Health Visiting*, *1*(4), 210-217.

Biddle, L., Paramasivan, S., Harris, S., Campbell, R., Brennan, J., & Hollingworth, W. (2016). Patients' and clinicians' experiences of holistic needs assessment using a cancer distress thermometer and problem list: a qualitative study. *European Journal of Oncology Nursing, 23*, 59-65.

Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in Psychology*, *3*(2), 77-101.

Braun, V., & Clarke, V. (2013). *Successful qualitative research: A practical guide for beginners*. London: Sage

Braun, V., & Clarke, V. (2019). Reflecting on reflexive thematic analysis. *Qualitative Research in Sport, Exercise and Health*, *11*(4), 589-597.

Braun, V., & Clarke, V. (2021a). Can I use TA? Should I use TA? Should I not use TA? Comparing reflexive thematic analysis and other pattern-based qualitative analytic approaches. *Counselling and Psychotherapy Research, 21*(1), 37-47.

Braun, V., & Clarke, V. (2021b). One size fits all? What counts as quality practice in (reflexive) thematic analysis?. *Qualitative Research in Psychology, 18*(3), 328-352.

Braun, V., & Clarke, V. (2021c). To saturate or not to saturate? Questioning data saturation as a useful concept for thematic analysis and sample-size rationales. *Qualitative Research in Sport, Exercise and Health*, *13*(2), 201-216.

Briggs-Gowan, M. J., & Carter, A. S. (2006). *BITSEA: Brief infant-toddler social and emotional assessment - Examiner's manual*. Harcourt Assessment.

Briggs-Gowan, M. J., Carter, A. S., McCarthy, K., Augustyn, M., Caronna, E., & Clark, R. (2013). Clinical validity of a brief measure of early childhood social–emotional/behavioural problems. *Journal of Paediatric Psychology*, *38*(5), 577-587.

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.

Chong, L. S., Fitzgerald, D. A., Craig, J. C., Manera, K. E., Hanson, C. S., Celermajer, D., ... & Tong, A. (2018). Children’s experiences of congenital heart disease: a systematic review of qualitative studies. *European Journal of Paediatrics, 177,* 319-336.

Clancy, T., Jordan, B., de Weerth, C., & Muscara, F. (2020). Early emotional, behavioural and social development of infants and young children with congenital heart disease: a systematic review. *Journal of Clinical Psychology in Medical Settings*, *27*(4), 686-703.

Dahlawi, N., Milnes, L. J., & Swallow, V. (2020). Behaviour and emotions of children and young people with congenital heart disease: A literature review. *Journal of Child Health Care*, *24*(2), 317-332.

De Buca, I., Maguire, C., & Ross, A. (2022). The views of non-clinical staff who participated in an Infant Mental Health (IMH) Training Day in a Child and Family service Cork, Ireland: A window into the general public perspective regarding infant mental health and wellbeing. *World Association for Infant Mental Health*. https://perspectives.waimh.org/2022/11/04/the-views-of-non-clinical-staff-who-participated-in-an-infant-mental-health-imh-training-day-in-a-child-and-family-service-cork-ireland-a-window-into-the-general-public-perspective-regarding-infan/

Ellis, A. (2017). *Relationships: parental mind-mindedness and infant outcomes, and how parents experience building a relationship with their baby on neonatal intensive care*. [DClinPsy Thesis, University of Birmingham]. https://etheses.bham.ac.uk/id/eprint/7761/1/Ellis17ClinPsyD_Vol_1.pdf

Fitzgerald, H. E., Weatherston, D., & Mann, T. L. (2011). Infant mental health: An interdisciplinary framework for early social and emotional development. *Current Problems in Paediatric and Adolescent Health Care, 41*(7), 178-182.

Gaskin, K. L. (2018). Patterns of transition experience for parents going home from hospital with their infant after first stage surgery for complex congenital heart disease. *Journal of Paediatric Nursing, 41*, e23-e32.

Gonzalez, V. J., Kimbro, R. T., Cutitta, K. E., Shabosky, J. C., Bilal, M. F., Penny, D. J., & Lopez, K. N. (2021). Mental health disorders in children with congenital heart disease. *Paediatrics*, *147*(2).

Gupta, S., Giuffre, R. M., Crawford, S., & Waters, J. (1998). Covert fears, anxiety and depression in congenital heart disease. *Cardiology in the Young, 8*(4), 491-499.

Harvey, K. A., Kovalesky, A., Woods, R. K., & Loan, L. A. (2013). Experiences of mothers of infants with congenital heart disease before, during, and after complex cardiac surgery. *Heart & Lung, 42*(6), 399-406.

Healthcare Improvement Scotland. (2019). *Congenital Heart Disease Standards.*

Hövels-Gürich, H. H., Konrad, K., Skorzenski, D., Minkenberg, R., Herpertz-Dahlmann, B., Messmer, B. J., & Seghaye, M. C. (2007). Long-term behaviour and quality of life after corrective cardiac surgery in infancy for tetralogy of Fallot or ventricular septal defect. *Paediatric Cardiology*, *28*(5), 346-354.

Hughes, K., Bellis, M. A., Hardcastle, K. A., Sethi, D., Butchart, A., Mikton, C., & Dunne, M. P. (2017). The effect of multiple adverse childhood experiences on health: a systematic review and meta-analysis. *The Lancet Public Health*, *2*(8), e356-e366.

Jordan, B., Franich-Ray, C., Anderson, V., Northam, E., Albert, N., Cochrane, A., & Menahem, S. (2013). Maternal report of infant emotional well-being following their infant's hospitalization for neonatal cardiac surgery. *IMH Journal*, *34*(4), 259-266.

Jordan, B., Franich-Ray, C., Albert, N., Anderson, V., Northam, E., Cochrane, A., & Menahem, S. (2014). Early mother-infant relationships after cardiac surgery in infancy. *Archives of Disease in Childhood, 99*(7), 641-645.

Kazak, A. E., Kassam-Adams, N., Schneider, S., Zelikovsky, N., Alderfer, M. A., & Rourke, M. (2006). An integrative model of paediatric medical traumatic stress. *Journal of Paediatric Psychology*, *31*(4), 343-355.

Kruizinga, I., Jansen, W., De Haan, C. L., Van der Ende, J., Carter, A. S., & Raat, H. (2012). Reliability and validity of the Dutch version of the Brief Infant-Toddler Social and Emotional Assessment (BITSEA). *PloS one*, *7*(6), e38762.

Larkin, M., Flowers, P., & Smith, J. A. (2021). Interpretative phenomenological analysis: Theory, method and research. *Interpretative Phenomenological Analysis,* 1-100.

Low, J. (2019). A pragmatic definition of the concept of theoretical saturation. *Sociological Focus*, *52*(2), 131-139.

Lumsden, M. R., Smith, D. M., & Wittkowski, A. (2019). Coping in parents of children with congenital heart disease: a systematic review and meta-synthesis. *Journal of Child and Family Studies, 28*, 1736-1753.

National Child Traumatic Stress Network (2003). *Paediatric Medical Traumatic Stress.* Retrieved 23 April, 2023 from www.nctsn.org/what-is-child-trauma/trauma-types/medical-trauma

Maslow, A. (1970). *Motivation and Personality.* New York: Harper & Row.

McMahon, E., & Chang, Y. S. (2020). From surviving to thriving-parental experiences of hospitalised infants with congenital heart disease undergoing cardiac surgery: A qualitative synthesis. *Journal of Paediatric Nursing*, *51*, 32-41.

McWhorter, L. G., Christofferson, J., Neely, T., Hildenbrand, A. K., Alderfer, M. A., Randall, A., & Sood, E. (2022). Parental post-traumatic stress, overprotective parenting, and emotional and behavioural problems for children with critical congenital heart disease. *Cardiology in the Young*, *32*(5), 738-745.

Meentken, M. G., Van Beynum, I. M., Legerstee, J. S., Helbing, W. A., & Utens, E. M. (2017). Medically related post-traumatic stress in children and adolescents with congenital heart defects*. Frontiers in Paediatrics, 5*, 20.

Meins, E., Fernyhough, C., Arnott, B., Leekam, S. R., & de Rosnay, M. (2013). Mind-mindedness and theory of mind: Mediating roles of language and perspectival symbolic play. *Child Development, 84*(5), 1777-1790.

Peters, J., Skirton, H., Morgan, J., & Clark, M. (2019). How do parents perceive and utilize knowledge of their infant's MH? A systematic review. *Journal of Child Health Care*, *23*(2), 242-255.

Rempel, G. R., Ravindran, V., Rogers, L. G., & Magill-Evans, J. (2013). Parenting under pressure: A grounded theory of parenting young children with life-threatening congenital heart disease. *Journal of Advanced Nursing*, *69*(3), 619-630.

Ryan, K. R., Jones, M. B., Allen, K. Y., Marino, B. S., Casey, F., Wernovsky, G., & Lisanti, A. J. (2019). Neurodevelopmental outcomes among children with congenital heart disease: at-risk populations and modifiable risk factors. *World Journal for Paediatric and Congenital Heart Surgery, 10*(6), 750-758.

Sameroff, A. J. (2000). Developmental systems and psychopathology. *Development and Psychopathology*, *12*(3), 297-312.

Silletti, F., Salvadori, E. A., Presaghi, F., Fasolo, M., Aureli, T., & Coppola, G. (2022). Maternal mind-mindedness during the first year of life: Developmental trajectories and moderators. *Developmental Psychology, 58*(9), 1615.

Sroufe, L. A. (2005). Attachment and development: A prospective, longitudinal study from birth to adulthood. *Attachment & Human Development*, *7*(4), 349-367.

Suttora, C., Spinelli, M., Aureli, T., Fasolo, M., Lionetti, F., Picciolini, O., ... & Salerni, N. (2020). Mind-mindedness and parenting stress: a cross-sectional study in a cohort of mothers of 3-month-old full-term and preterm infants. *International Journal of Environmental Research and Public Health*, *17*(21), 7735.

Szaniecki, E., & Barnes, J. (2016). Measurement issues: Measures of IMH. *Child and Adolescent MH*, *21*(1), 64-74.

The Scottish Government. (2020). *Evaluability assessment of the Scottish Government's Perinatal and IMH Programme.* https://www.gov.scot/binaries/content/documents/govscot/publications/research-and-analysis/2021/03/evaluability-assessment-scottish-governments-perinatal-infant-mental-health-programme/documents/evaluability-assessment-scottish-governments-perinatal-infant-mental-health-programme-2020/evaluability-assessment-scottish-governments-perinatal-infant-mental-health-programme-2020/govscot%3Adocument/evaluability-assessment-scottish-governments-perinatal-infant-mental-health-programme-2020.pdf

Tesson, S., Butow, P. N., Marshall, K., Fonagy, P., & Kasparian, N. A. (2022). Parent-child bonding and attachment during pregnancy and early childhood following congenital heart disease diagnosis. *Health Psychology Review, 16*(3), 378-411.

Tong, A., Sainsbury, P., & Craig, J. (2007). Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care, 19*(6), 349-357.

Wei, H., Roscigno, C. I., & Swanson, K. M. (2017). Healthcare providers' caring: Nothing is too small for parents and children hospitalized for heart surgery. *Heart & Lung*, *46*(3), 166-171.

Woolf-King, S. E., Anger, A., Arnold, E. A., Weiss, S. J., & Teitel, D. (2017). Mental health among parents of children with critical congenital heart defects: a systematic review. *Journal of the American Heart Association, 6*(2), e004862.

Appendices

Appendix A

Completed Reporting Guidelines - ENTREQ

No. Item	Guide questions/description	Reported on Page #
1. Aim	State the research question the synthesis addresses	18
2. Synthesis methodology	Identify the synthesis methodology or theoretical framework which underpins the synthesis, and describe the rationale for choice of methodology (e.g. meta-ethnography, thematic synthesis, critical interpretive synthesis, grounded theory synthesis, realist synthesis, meta-aggregation, meta-study, framework synthesis)	18
3. Approach to searching	Indicate whether the search was pre-planned (comprehensive search strategies to seek all available studies) or iterative (to seek all available concepts until they theoretical saturation is achieved)	18
4. Inclusion criteria	Specify the inclusion/exclusion criteria (e.g. in terms of population, language, year limits, type of publication, study type)	18-19
5. Data sources	Describe the information sources used (e.g. electronic databases (MEDLINE, EMBASE, CINAHL, psycINFO), grey literature databases (digital thesis, policy reports), relevant organisational websites, experts, information specialists, generic web searches (Google Scholar) hand searching, reference lists) and when the searches conducted; provide the rationale for using the data sources	20
6. Electronic Search strategy	Describe the literature search (e.g. provide electronic search strategies with population terms, clinical or health topic terms, experiential or social phenomena related terms, filters for qualitative research, and search limits)	19
7. Study screening methods	Describe the process of study screening and sifting (e.g. title, abstract and full text review, number of independent reviewers who screened studies)	20-21
8. Study characteristics	Present the characteristics of the included studies (e.g. year of publication, country, population, number of participants, data collection, methodology, analysis, research questions)	28-35
9. Study selection results	Identify the number of studies screened and provide reasons for study exclusion (e,g, for comprehensive searching, provide numbers of studies screened and reasons for exclusion indicated in a figure/flowchart; for iterative searching describe reasons for study exclusion and inclusion based on modifications to the	22

	research question and/or contribution to theory development)	
10. Rationale for appraisal	Describe the rationale and approach used to appraise the included studies or selected findings (e.g. assessment of conduct (validity and robustness), assessment of reporting (transparency), assessment of content and utility of the findings)	23
11. Appraisal items	State the tools, frameworks and criteria used to appraise the studies or selected findings (e.g. Existing tools: CASP, QARI, COREQ, Mays and Pope [25]; reviewer developed tools; describe the domains assessed: research team, study design, data analysis and interpretations, reporting)	23
12. Appraisal process	Indicate whether the appraisal was conducted independently by more than one reviewer and if consensus was required	23
13. Appraisal results	Present results of the quality assessment and indicate which articles, if any, were weighted/excluded based on the assessment and give the rationale	38
14. Data extraction	Indicate which sections of the primary studies were analysed and how were the data extracted from the primary studies? (e.g. all text under the headings “results /conclusions” were extracted electronically and entered into a computer software)	23
15. Software	State the computer software used, if any	23
16.Number of reviewers	Identify who was involved in coding and analysis	24
17. Coding	Describe the process for coding of data (e.g. line by line coding to search for concepts)	24
18. Study comparison	Describe how were comparisons made within and across studies (e.g. subsequent studies were coded into pre-existing concepts, and new concepts were created when deemed necessary)	24
19. Derivation of themes	Explain whether the process of deriving the themes or constructs was inductive or deductive	24
20.Quotations	Provide quotations from the primary studies to illustrate themes/constructs, and identify whether the quotations were participant quotations of the author’s interpretation	41-47
21. Synthesis output	Present rich, compelling and useful results that go beyond a summary of the primary studies (e.g. new interpretation, models of evidence, conceptual models, analytical framework, development of a new theory or construct)	40

Appendix B

Example of Search Strategy in CINAHL, PsychInfo, & Medline

Interface: EBSCOhost

Database: CINAHL

Date searched: 27.06.22

Retrieved records: 918

#	Search strategy	Results
S72	S5 AND S9 AND S70; NARROW BY LANGUAGE – ENGLISH	918
S71	S5 AND S9 AND S70	926
S70	S32 OR S69	1,656,403
S69	S33 OR S34 OR S35 OR S36 OR S37 OR S38 OR S39 OR S40 OR S41 OR S42 OR S43 OR S44 OR S45 OR S46 OR S47 OR S48 OR S49 OR S50 OR S51 OR S52 OR S53 OR S54 OR S55 OR S56 OR S57 OR S58 OR S59 OR S60 OR S61 OR S62 OR S63 OR S64 OR S65 OR S66 OR S67 OR S68	1,377,949
S68	TX "narrative analys#s"	1,458
S67	TX constant n3 (comparative or comparison)	10,265
S66	TI ((discourse* or discurs*) n3 analys#s) OR AB ((discourse* or discurs*) n3 analys#s)	2,965
S65	TX "discourse analys#s"	5,969
S64	TX "constant comparative"	9,255
S63	TX "thematic analysis"	82,737
S62	TX "content analysis"	49,626
S61	TX questionnaire*	598,704
S60	TX focus group*	62,372
S59	TX field stud*	15,415
S58	TX observational method*	28,417
S57	TX categor*	151,681
S56	TX theme* or thematic	141,030

S55	TX "cluster sampl*"	7,129
S54	TX "life experience*"	36,832
S53	TI "lived experience*" OR AB "lived experience*"	9,579
S52	TX "life world" or life-world or "conversation analys#s" or "personal experience*" or "theoretical saturation"	7,525
S51	TX interview*	390,427
S50	TX account or accounts or unstructured or open-ended or "open ended" or text* or narrative*	218,206
S49	TX (purpos* n4 sampl*) or (focus n2 group*)	95,798
S48	TX "theoretical sampl*"	2,328
S47	TX "purposive sampl*"	38,358
S46	TX "qualitative validity"	563
S45	TI "biographical method" OR AB "biographical method"	15
S44	TI "human science" OR AB "human science"	222
S43	TI (field n2 (study or studies or research)) OR AB (field n2 (study or studies or research))	12,454
S42	TX (humanistic or existential or experiential or paradigm*)	50,129
S41	TX "action research" OR "co operative inquir*" OR "cooperative inquir*" OR "co-operative inquir*"	11,483
S40	TX social construct* OR (postmodern* or post-structural*) OR ("post structural*" or poststructural*) OR "post modern*" OR (post-modern* or interpret*)	117,774
S39	TX (emic or etic or hermeneutic* or heuristic* or semiotic*) OR TI data n2 saturat* AND AU data n2 saturat* OR TI "participant observ*" AND AB "participant observ*"	8,333
S38	TX "life stor*"	1,389
S37	TX (grounded n2 (theor* or study or studies or research or analys#s))	21,942
S36	TX grounded theory	21,313
S35	TX phenomenol*	27,462
S34	TX ethnograph*	14,467

S33	TX ethnonursing	315
S32	S10 OR S11 OR S12 OR S13 OR S14 OR S15 OR S16 OR S17 OR S18 OR S19 OR S20 OR S21 OR S22 OR S23 OR S24 OR S25 OR S26 OR S27 OR S28 OR S29 OR S30 OR S31	1,073,297
S31	(MH "Cluster Sample+")	5,905
S30	(MH "Life Experiences+")	50,403
S29	(MH "Phenomenological Research")	17,770
S28	(MH "Phenomenology")	3,995
S27	(MH "Theoretical Sample")	1,855
S26	(MH "Field Studies")	3,506
S25	(MH "Observational Methods+")	21,857
S24	(MH "Purposive Sample")	35,428
S23	(MH "Qualitative Validity+")	1,759
S22	(MH "Constant Comparative Method")	7,457
S21	(MH "Ethnonursing Research")	219
S20	(MH "Ethnological Research")	6,810
S19	(MH "Ethnographic Research")	8,744
S18	(MH "Content Analysis")	40,395
S17	(MH "Thematic Analysis")	75,786
S16	(MH "Interviews+")	240,629
S15	(MH "Discourse Analysis")	5,300
S14	(MH "Focus Groups")	48,310
S13	(MH "Attitude+")	501,663
S12	(MH "Questionnaires+")	460,403
S11	(MH "Research, Nursing")	22,186
S10	(MH "Qualitative Studies")	131,984
3rd Concept		
S9	S6 OR S7 OR S8	1,128,932

S8	TI (child* OR adolescen* OR teen* OR young pe* OR juvenile OR youth) OR AB (child* OR adolescen* OR teen* OR young pe* OR juvenile OR youth)	703,381
S7	(MH "Adolescence")	576,534
S6	(MH "Child")	506267
2nd concept		
S5	S1 OR S2 OR S3 OR S4	22785
S4	TI (inflammatory bowel disease OR crohn* OR ulcerative colitis) OR AB (inflammatory bowel disease OR crohn* OR ulcerative colitis)	18830
S3	(MH "Colitis, Ulcerative")	5869
S2	(MH “Crohn Disease”)	7624
S1	(MH “Inflammatory Bowel Diseases”)	8147
1st Concept		

Interface: EBSCOhost

Database: PsychINFO

Date searched: 27.06.22

Retrieved records: 198

#	Query	Results
S65	S5 AND S10 AND S63; NARROW BY LANGUAGE - ENGLISH	198
S64	S5 AND S10 AND S63	210
S63	S25 OR S62	1932491
S62	S26 OR S27 OR S28 OR S29 OR S30 OR S31 OR S32 OR S33 OR S34 OR S35 OR S36 OR S37 OR S38 OR S39 OR S40 OR S41 OR S42 OR S43 OR S44 OR S45 OR S46 OR S47 OR S48 OR S49 OR S50 OR S51 OR S52 OR S53 OR S54 OR S55 OR S56 OR S57 OR S58 OR S59 OR S60 OR S61	1895048
S61	TX "narrative analys#s"	3266
S60	TX constant n3 (comparative or comparison)	5569
S59	TI ((discourse* or discurs*) n3 analys#s) OR AB ((discourse* or discurs*) n3 analys#s)	9392

S58	TX "discourse analys#s"	13989
S57	TX "constant comparative"	3735
S56	TX "thematic analysis"	20864
S55	TX "content analysis"	32249
S54	TX questionnaire*	520298
S53	TX focus group*	60771
S52	TX field stud*	57986
S51	TX observational method*	5744
S50	TX categor*	188168
S49	TX theme* or thematic	160018
S48	TX "cluster sampl*"	1869
S47	TX "life experience*"	37564
S46	TI "lived experience*" OR AB "lived experience*"	18519
S45	TX "life world" or life-world or "conversation analys#s" or "personal experience*" or "theoretical saturation"	30889
S44	TX interview*	587168
S43	TX account or accounts or unstructured or open-ended or "open ended" or text* or narrative*	399202
S42	TX (purpos* n4 sampl*) or (focus n2 group*)	58002
S41	TX "theoretical sampl*"	666
S40	TX "purposive sampl*"	6313
S39	TX "qualitative validity"	11
S38	TI "biographical method" OR AB "biographical method"	59
S37	TI "human science" OR AB "human science"	601
S36	TI (field n2 (study or studies or research)) OR AB (field n2 (study or studies or research))	38054
S35	TX (humanistic or existential or experiential or paradigm*)	158,676
S34	TX "action research" OR "co operative inquir*" OR "cooperative inquir*" OR "co-operative inquir*"	11553

S33	TX social construct* OR (postmodern* or post-structural*) OR ("post structural*" or poststructural*) OR "post modern*" OR (post-modern* or interpret*)	259154
S32	TX (emic or etic or hermeneutic* or heuristic* or semiotic*) OR TI data n2 saturat* AND AU data n2 saturat* OR TI "participant observ*" AND AB "participant observ*"	31022
S31	TX "life stor*"	4,226
S30	TX (grounded n2 (theor* or study or studies or research or analys#s))	24,256
S29	TX grounded theory	22,239
S28	TX phenomenol*	52171
S27	TX ethnograph*	33288
S26	TX ethnonursing	66
S25	S11 OR S12 OR S13 OR S14 OR S15 OR S16 OR S17 OR S18 OR S19 OR S20 OR S21 OR S22 OR S23 OR S24	183774
S24	DE "Life Experiences" OR DE "Life Changes"	32865
S23	DE "Phenomenology" OR DE "Interpretative Phenomenological Analysis"	17456
S22	DE "Observation Methods"	5944
S21	DE "Narrative Analysis"	1053
S20	DE "Ethnology" OR DE "Ethnography"	12411
S19	DE "Content Analysis"	5742
S18	DE "Thematic Analysis"	1633
S17	DE "Interviews" OR DE "Intake Interview" OR DE "Interview Schedules" OR DE "Psychodiagnostic Interview" OR DE "Semi-Structured Interview"	15857
S16	DE "Discourse Analysis"	9532
S15	DE "Focus Group" OR DE "Focus Group Interview"	1020
S14	DE "Attitudes"	29732
S13	DE "Questionnaires" OR DE "General Health Questionnaire"	23869
S12	DE "Nursing"	25808
S11	DE "Qualitative Methods"	10511
3rd Concept		

S10	S6 OR S7 OR S8 OR S9	1,015,120
S9	TI (child* OR adolescen* OR teen* OR young pe* OR juvenile OR youth) OR AB (child* OR adolescen* OR teen* OR young pe* OR juvenile OR youth)	982846
S8	DE adolescents	0
S7	DE Adolescence	0
S6	DE Child	168578
2nd Concept		
S5	S1 OR S2 OR S3 OR S4	1827
S4	TI (inflammatory bowel disease OR crohn* OR ulcerative colitis) OR AB (inflammatory bowel disease OR crohn* OR ulcerative colitis)	1797
S3	DE Crohns disease	0
S2	DE "Ulcerative Colitis"	309
S1	DE inflammatory bowel disease	0
1st Concept		

Interface: EBSCOhost **Database: Medline** **Date searched: 27.06.22** **Retrieved records: 2506**		
#	**Query**	**Results**
S69	S5 AND S9 AND S67: Narrow by language - English	2,506
S68	S5 AND S9 AND S67	2,658
S67	S65 OR S66	3,142,148
S66	S10 OR S11 OR S12 OR S13 OR S14 OR S15 OR S16 OR S17 OR S18 OR S19 OR S20 OR S21 OR S22 OR S23 OR S24 OR S25 OR S26 OR S27 OR S28 OR S29	327,343
S65	S30 OR S31 OR S32 OR S33 OR S34 OR S35 OR S36 OR S37 OR S38 OR S39 OR S40 OR S41 OR S42 OR S43 OR S44 OR S45 OR S46 OR S47 OR S48 OR S49 OR S50 OR S51 OR S52 OR S53 OR S54 OR S55 OR S56 OR S57 OR S58 OR S59 OR S60 OR S61 OR S62 OR S63 OR S64	3090688

S64	TX (narrative analys#s)	6093
S63	TX (constant n2 (comparative or comparison)) ,	6477
S62	AB (((discourse* or discurs*) n4 analys#s)) OR TI (((discourse* or discurs*) n4 analys#s))	3040
S61	TX (discourse analys#s)	3657
S60	TX (constant comparative)	3657
S59	TX (thematic analysis)	40506
S58	TX (content analysis)	66680
S57	TX questionnaire*	896258
S56	TX (focus group*)	73273
S55	TX field stud*	79292
S54	TX (observational method*)	7728
S53	TX categor*	448790
S52	TX (theme* or thematic)	161654
S51	TX (cluster sampl*)	16064
S50	TX life experience*	27748
S49	AB (lived experience*) OR TI (lived experience*)	9536
S48	TX ((life world) or life-world or conversation analys#s or personal experience* or theoretical saturation)	33168
S47	TX (account or accounts or unstructured or open-ended or (open ended) or text* or narrative*	814213
S46	TX ((purpos* n5 sampl*) or (focus adj group*))	21851
S45	TX (theoretical sampl*)	2370
S44	TX (purposive sampl*)	11955
S43	TX "qualitative validity"	24
S42	AB (biographical method) OR TI (biographical method)	78
S41	AB (human science) OR TI (human science)	3997
S40	AB ((field n2 (study or studies or research)) OR TI ((field n2 (study or studies or research))	74005

S39	TX (humanistic or existential or experiential or paradigm*)	206083
S38	TX (action research or cooperative inquir* or (co operative inquir*) or (co-operative inquir*))	17137
S37	TX ((social construct* or (postmodern* or post-structural*) or (post structural* or poststructural*) or (post modern*) or post-modern* or feminis* or interpret*)	587, 814
S36	TX ((emic or etic or hermeneutic* or heuristic* or semiotic*)) OR AB ((data n2 saturat*) or (participant observ*)) OR TI ((data n2 saturat*) or (participant observ*))	45857
S35	TX ((life stor*) or (women's stor*))	7508
S34	TX (grounded n2 (theor* or study or studies or research or analys#s))	18442
S33	TX grounded theory	17628
S32	TX phenomenol*	35537
S31	TX ethnograph*	15559
S30	TX ethnonursing	141
S29	TX "cluster sampl*"	8772
S28	TX "life experience*"	8123
S27	TX phenomenological research	1791
S26	MH "phenomenology"	70
S25	TX theoretical sampl*	2370
S24	TX field stud*	79292
S23	TX observational method*	7728
S22	TX purposive sample	4748
S21	TX qualitative validity	1023
S20	TX constant comparative method	2223
S19	TX ethnonursing research	61
S18	TX ethnological research	24
S17	TX ethnographic research	1800
S16	TX content analysis	66,680

S15	TX discourse analysis	3647
S14	MH "Focus Groups"	34317
S13	MH "Attitude"	51,743
S12	MH "Questionnaires"	6348
S11	MH "Nursing Methodology Research"	16406
S10	(MH "Qualitative Research")	74300
3rd Concept		
S9	S6 OR S7 OR S8	3,649,197
S8	AB (child* OR adolescen* OR teen* OR young pe* OR juvenile OR youth) OR TI (child* OR adolescen* OR teen* OR young pe* OR juvenile OR youth)	1,496,094
S7	(MH "Adolescent")	2,174,781
S6	(MH "Child")	1842889
2nd Concept		
S5	S1 OR S2 OR S3 OR S4	123,617
S4	AB (inflammatory bowel disease OR crohn* OR ulcerative colitis) OR TI (inflammatory bowel disease OR crohn* OR ulcerative colitis)	89,766
S3	(MH "Colitis, Ulcerative")	38448
S2	(MH "Crohn Disease")	42579
S1	(MH "Inflammatory Bowel Diseases")	27746
1st Concept		

Appendix C

Joanna Briggs Institute Critical Appraisal Checklist for Qualitative Studies

JBI CRITICAL APPRAISAL CHECKLIST FOR QUALITATIVE RESEARCH

Reviewer __________________ Date _________

Author _________________ Year________ Record Number_________

	Yes	No	Unclear	Not applicable
1. Is there congruity between the stated philosophical perspective and the research methodology?	☐	☐	☐	☐
2. Is there congruity between the research methodology and the research question or objectives?	☐	☐	☐	☐
3. Is there congruity between the research methodology and the methods used to collect data?	☐	☐	☐	☐
4. Is there congruity between the research methodology and the representation and analysis of data?	☐	☐	☐	☐
5. Is there congruity between the research methodology and the interpretation of results?	☐	☐	☐	☐
6. Is there a statement locating the researcher culturally or theoretically?	☐	☐	☐	☐
7. Is the influence of the researcher on the research, and vice- versa, addressed?	☐	☐	☐	☐
8. Are participants, and their voices, adequately represented?	☐	☐	☐	☐
9. Is the research ethical according to current criteria or, for recent studies, and is there evidence of ethical approval by an appropriate body?	☐	☐	☐	☐
10. Do the conclusions drawn in the research report flow from the analysis, or interpretation, of the data?	☐	☐	☐	☐

Overall appraisal: Include ☐ Exclude ☐ Seek further info ☐

Comments (Including reason for exclusion)

____

____

____

Appendix D

Sample of Thematic Synthesis Reflexivity Log

Beliefs, knowledge, experiences and understandings of lead reviewer that likely impacted interpretation:

1. A core training in psychopathology
    - Potentially prone to looking for risk/vulnerability factors and being drawn to emotional and social challenges that CYP in the studies describe (attentional biases)

2. Clinical experience predominantly in CAMHS
    - Inevitably adopt systemic lens, aligned to ecological systems theory, thinking about the multiple environments and contexts of the CYP as the primary source of their experiences.
    - My beliefs/assumptions within this: families/caregivers mediate a child's contact with the wider world and themselves
    - From limited clinical experience of working with CYP with IBD found those around the child/YP play a key, influential role in fostering adjustment process, resilience, & acceptance

3. Clinical preference for third wave approaches, such as ACT or CFT, that more actively use metaphors and visualisation to aid understanding
    - Noticed this shaped my reading of data and interpretations, for example: in interpreting personifications in the data of IBD as an unwanted guest (aligned with concepts from ACT)
    - Also attended to self-criticism and shame within participants excerpt, likely from use of CFT

Appendix E

Coding Excerpt

Reference	Data from Original Articles	Codes
Adolescents lived experiences while hospitalised after surgery for UC Olsen et al., (2016)	Body: Out of Order The adolescents described how the physical and mental challenges had brought them to the end of their tether: “If I really had to say what I thought at the time, it’s that ‘now it’s over, I won’t survive this, and that’s probably for the best’” (Informant 2). They indicated that they had been well informed before the first operation and had felt well prepared. But despite their previous experiences with a body that was “out of order” and had failed them through the many periods of illness before the operation, they had been surprised by the body’s reactions in the postoperative course. Some put it this way, “You actually don’t know what is going on inside your body” (Informants 6 and 8). The adolescents gave blunt descriptions of their physical condition; for example, “It was just bloody awful” (Informants 4 and 6). Their remarks reflected an image of a body beyond control: “How can you keep throwing up, in huge quantities, and for several days, when you’re not eating anything?” (Informant 6). The nausea was experienced as paralyzing (Informant 4), hindering them from being mobilized, taking food, or even entering into conversation. One informant verbalized a sense of shock and anxiety, “I was nauseous and shaking ... I was shaking like mad. I just felt so sick” (Informant 7). There was a feeling that it would never stop. As if with one voice, they said, “I felt really awful,” another frequent formulation being, “I felt absolutely rotten.”	Physical and MH difficulties Hopelessness about future Well-prepared for surgery Well-prepared for surgery Body as distinct and defunct (hence need for surgery) Uncertainty with inner workings of body Helpless to severe symptoms Disbelief at severity of symptoms Helpless to severe symptoms Helpless to severe symptoms Strong emotional response post-surgery Relentless impact of IBD Helpless to severe symptoms
	A temporary ostomy was performed as part of the series of operations. On the one hand, the ostomy was referred to as “gross” (Informants 1, 6, and 7), on the other hand, an informant said, “I thought it would be harder than it actually was—you know, it sounds so dramatic having the small intestine coming out of your stomach—you have these screwed up ideas (Informant 5).	Disgust about ostomy Reality not as bad as expectations Self-criticism
	A distancing attitude to the ostomy was also evidenced by the use of metaphors such as “It was kind of like borrowing a dress from a friend” (Informant 6). When the ostomy began functioning properly, the adolescents were quick to learn how to cut from the template, clean the appliance, and change the bag. One informant put it like this, “After a little while, when I	Ostomy as separate to self Quickly adapted to demands of ostomy

had learned to put it on myself, you know, I got the knack of cutting a hole in that sticky thing there that you have to put on” (Informant 1).	Quickly adapted to demands of ostomy
The adolescents described how the exhaustion left them drained of energy and initiative, “I couldn’t even swallow a paracetamol” (Informant 5), and, “I said ‘but I can’t, I can’t’—I simply could not do it” (Informant 8). Besides experiencing physical exhaustion they felt mentally weakened, “I was vomiting and feeling really ill—I could hardly face having my family visiting ... all they could do was to just sit there and watch me throw up all over the place” (Informant 5). Their exhaustion was accompanied by a feeling that they would not survive, or thoughts of suicide:	Mastery over ostomy Wider physical impacts of procedure Helpless and exhausted Helpless to severe symptoms
“If I had been able to, I’d have jumped out of that window there. I really would. I was totally incapable of seeing how I could face the future. Since then, I haven’t had suicidal thoughts, but that night I’d just had it.” (Informant 6)	Suicidal ideation Suicidal ideation Hopelessness about future
Suicidal thoughts were seen as inescapable:	
There was several times when I said, “Let me just have the final jab and then it’s over, because I can’t take any more.” I did not want to be here any longer because I felt so bad and it just hurt. (Informant 7)	Helpless to mental impacts of procedure Suicidal ideation
The adolescents did not share their thoughts about death with parents or nurses: “Those things you can- not talk about” (Informants 4) and “I could sense that it was hard on my parents ... so maybe [dying] would just be the easiest thing” (Informant 4).	Hopelessness about future Suppression of thoughts & feelings Feeling like a burden
The emotional chaos gave rise to a need for deep rest, and their single rooms allowed them to close out the world and leave communication and other matters to the parents: “So I’d close my ears thinking, ‘Mum will be hearing that’” (Informant 5).	Helpless and exhausted Give parents/others responsibility
Seen and Understood	
The adolescents gave accounts of the several lengthy illness episodes they had undergone before the operation. Their experiences had had a profound effect on school and their social and family life: “I have done my best to delete all the images from that time” (Informant 4). They spoke about ups and downs according to whether the medical treatment had been successful or not. They had experienced constantly recurring stomach pain, bloody diarrheas, side effects causing massive weight gain, changes in facial form, and acne: “Some of my cousins came over to me [at a social function] and introduced themselves, because they were unable to recognize me—they had no idea who they were talking to. They thought I was somebody’s girlfriend” (Informant 7). Some had felt rejected by their doctor’s remarks that their stomach problems were “just a mental thing” (Informant 7). Outside of the hospital world,	Validation from others A difficult journey leading up to procedure Ripple effects on system Avoidance Wellbeing seen as contingent on medical treatment effectiveness (external locus of control?) Severe symptoms

	the adolescents had also felt deserted, misunderstood, and ignored: “By the end of year 9 [aged 15] I had no friends left. I was placed at a single desk, because, as the others said, I was never there anyway, so why should they bother sitting next to me” (Informant 7). This made it even more important that their parents were admitted with them. The parents’ ability to see through the illness and suffering enabled them to offer protection and to act as their guardians when the situation became too difficult for them. This was indirectly expressed in the remark, “I know what a bloody nuisance one’s parents can be, but after all, they do know how their child is doing, and how they prefer things to be done, so I was extremely happy that they were with me” (Informant 4). The adolescents did not directly question the nurses’ technical instrumental skills, for example, whether they were heavy-handed, or did too much or too little in relation to ostomy, catheter, or wound care, and so forth. They did distinguish, however, between those nurses they said were “nice” or “fantastic” and those they called “surly” or “cold.” Being nice or fantastic related to the nurse’s ability to meet and understand the individual person: “She was so nice and understood everything—about friends, interests and that sort of thing. She was cheerful and positive, and able to put herself in my place” (Informant 4). The ability to appear calm and take the necessary time was emphasized: “It was absolutely as if for her, it didn’t matter how long it took” (Informant 1). It was clear that the meeting with the nurses also had other dimensions: “What I liked was that they would take the initiative. ... [It was] as good as speaking to a psychologist. ... They actually cared and didn’t just do it because they had to” (Informant 1). The presence of nurses with these qualities was seen as particularly important: “When they were there, I did not have the same need for my parents to be there” (Informant 4). The adolescents stressed the significance of being seen and taken seriously, and when this happened, the nurse was seen as someone who respected them and tried to understand the extent and importance of what they were going through: “That was the impression they gave me, anyway—that it mattered to them who took care of me, and that I shouldn’t be seeing too many different nurses, and we had sort of bonded with each other, and that they would also like to build on that.” (Informant 4) Nurses’ surliness or coldness was associated with various dismissive kinds of behavior, as indicated by remarks such as the following: “Some of them were very stiff. There was no way you could swing with them at all” (Informant 8), or “But those cross and cold nurses ... it was mostly the older ones ... they’d say ‘There, it’s like this and like that” (Informant 7). Interaction with these nurses became strained when the adolescents felt that nothing was negotiable and that they were neither being heard nor understood: “She was just so cold ... she had no feelings whatsoever” (Informant 4). This would happen when they felt that the nurses pressed	Impact on body image & appearance- completely changed by symptoms Invalidation from health professionals Isolated The self as invisible Parents unconditional support Parents as important advocates Parents support as crucial to coping Accepting of health interventions Different nursing styles Nurses considerate of the YP as a whole, not just their medical needs Empathic nurses Not feeling rushed as helpful Nurses not only provide physical/medical support Gratitude for nurses care Nurses as substitute/secondary attachment figure Person-centred care Attachment with nursing staff Lack of compassionate care remembered Rigidity from care providers Feeling invisible Tension in the air with nursing staff Different aims from care providers

them beyond their limits and in ways that had no appeal for them. The result was a distance between the adolescent, the nurse, and what should have been their common goal of healing: “I think she was very domineering ... ‘Now we do this’ and ‘Now we do that’, you know” (Informant 1). It seems that the adolescent patients were occasionally pressed beyond control: “I simply gave her such a mouthful” (Informant 8).

Interaction with the nurses took many different forms. A humorous approach was often emphasized as a redeeming feature of a chaotic time, “Maybe they poked fun at me if I was a bit cross, but that would just cheer me up” (Informant 8). The importance of the tone was stressed, however, “‘In a soft way, please’— not the rough way, you know” (Informant 6). Even when a more serious tone was used, it was acknowledged to have the effect of stimulating cooperation, “...like a well-meant verbal kick in the backside” (Informant 5). In heart-to-heart conversations, the adolescents were given opportunity to discuss their worries and other problems that they avoided sharing with their parents.

Where Are All the Others?

Surgery was conducted in a specialized unit that admitted both adults and adolescents, and the adolescents expressed a feeling of being out of place, “Oh no, no, no—those old people, I just can’t face that” (Informant 5). To them, the age difference obstructed the possibility of social interaction: “You don’t have a lot in common with them [the adult copatients]” (Informant 2). Having to be with and look at the elderly copatients was distasteful to the adolescents: “They were all ill people, old ladies, who I didn’t fancy looking at” (Informant 3). The adolescents missed the company of peers. Their longing for a glimpse of other young people in the corridors was frequently voiced. This would have created recognition and camaraderie, they felt:

> Then you would get out of your single room, because that’s a hell of a bore after a while. There’s no socializing there, you know. Let’s say there was a [young patient] further along the corridor. So the two of us could sit down together in the visiting room and watch TV and have a snack—we might have had a bit of fun then. (Informant 5)

Before the operation, the adolescents were offered contact with or a visit from a young person who had undergone the procedure. They had all declined the offer, however, worrying that they might hear about bad experiences. So, despite their sense of being well informed before the operation, they were in fact totally unprepared for how their body would be affected. They realized that learning from a peer’s experiences might have helped them start eating again,

Rigidity from care providers/Feeling invisible
Pushed to their limits by care providers
Others remaining light in face of turmoil

Accepting encouragement from nurses, even if it wasn’t easy
Nurses as substitute/secondary attachment figure

Lack of connection with similar others in hospital
Lack of connection with similar others in hospital
Feeling different to older IBD patients

Feeling isolated and alone

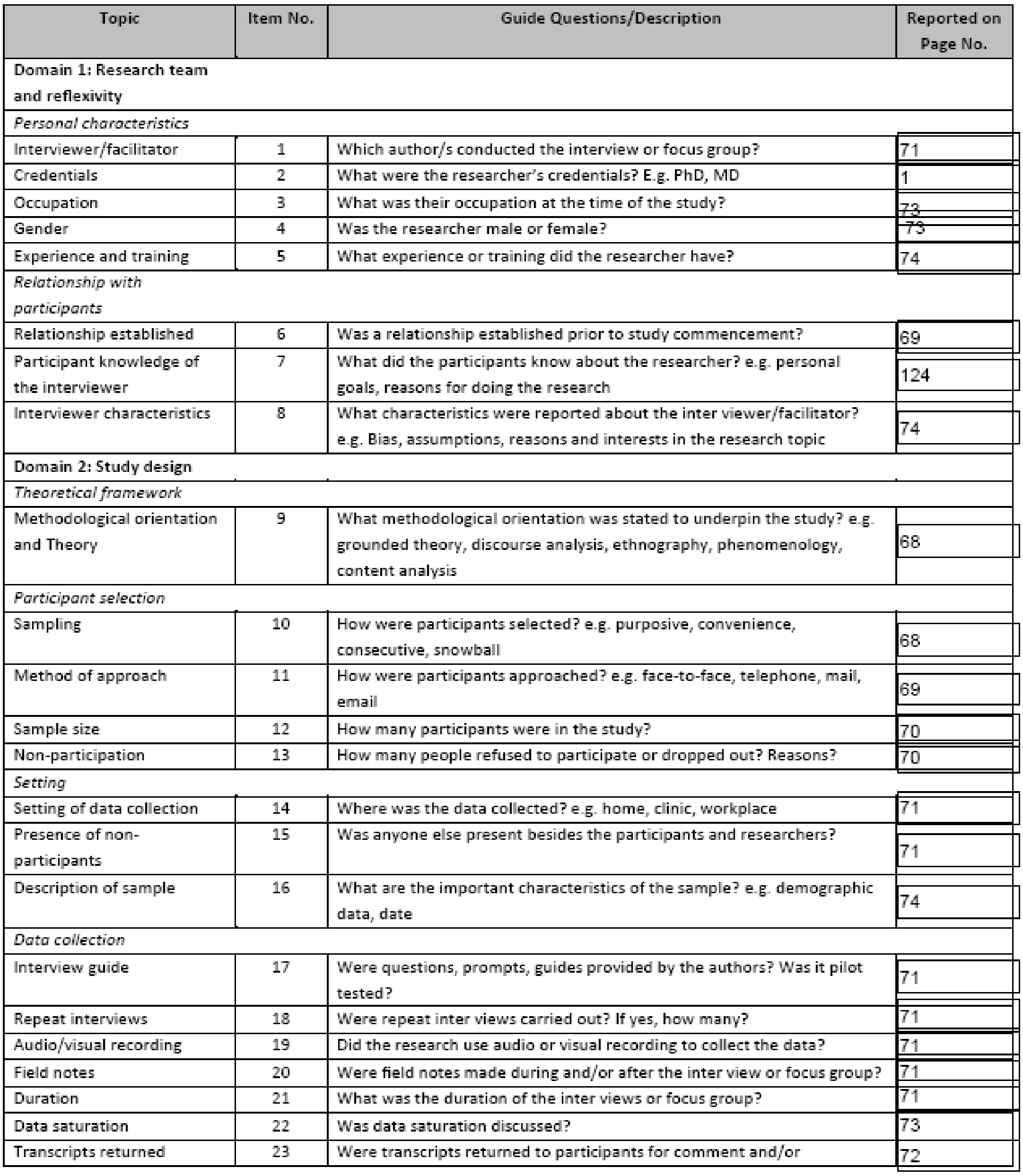
Feeling isolated and alone
Wishing for a different experience
Avoidance due to anxiety

Regretful of previous avoidance

	getting mobilized, and understand that their situation was nothing out of the ordinary: “Yeah, [talking to] some- one who had been a patient like me—I missed that” (Informant 7).	

Appendix F

Completed COREQ Checklist



Topic	Item No.	Guide Questions/Description	Reported on Page No.
Domain 1: Research team and reflexivity			
Personal characteristics			
Interviewer/facilitator	1	Which author/s conducted the interview or focus group?	71
Credentials	2	What were the researcher's credentials? E.g. PhD, MD	1
Occupation	3	What was their occupation at the time of the study?	73
Gender	4	Was the researcher male or female?	73
Experience and training	5	What experience or training did the researcher have?	74
Relationship with participants			
Relationship established	6	Was a relationship established prior to study commencement?	69
Participant knowledge of the interviewer	7	What did the participants know about the researcher? e.g. personal goals, reasons for doing the research	124
Interviewer characteristics	8	What characteristics were reported about the inter viewer/facilitator? e.g. Bias, assumptions, reasons and interests in the research topic	74
Domain 2: Study design			
Theoretical framework			
Methodological orientation and Theory	9	What methodological orientation was stated to underpin the study? e.g. grounded theory, discourse analysis, ethnography, phenomenology, content analysis	68
Participant selection			
Sampling	10	How were participants selected? e.g. purposive, convenience, consecutive, snowball	68
Method of approach	11	How were participants approached? e.g. face-to-face, telephone, mail, email	69
Sample size	12	How many participants were in the study?	70
Non-participation	13	How many people refused to participate or dropped out? Reasons?	70
Setting			
Setting of data collection	14	Where was the data collected? e.g. home, clinic, workplace	71
Presence of non-participants	15	Was anyone else present besides the participants and researchers?	71
Description of sample	16	What are the important characteristics of the sample? e.g. demographic data, date	74
Data collection			
Interview guide	17	Were questions, prompts, guides provided by the authors? Was it pilot tested?	71
Repeat interviews	18	Were repeat inter views carried out? If yes, how many?	71
Audio/visual recording	19	Did the research use audio or visual recording to collect the data?	71
Field notes	20	Were field notes made during and/or after the inter view or focus group?	71
Duration	21	What was the duration of the inter views or focus group?	71
Data saturation	22	Was data saturation discussed?	73
Transcripts returned	23	Were transcripts returned to participants for comment and/or	72

Topic	Item No.	Guide Questions/Description	Reported on Page No.
		correction?	
Domain 3: analysis and findings			
Data analysis			
Number of data coders	24	How many data coders coded the data?	73
Description of the coding tree	25	Did authors provide a description of the coding tree?	76
Derivation of themes	26	Were themes identified in advance or derived from the data?	72
Software	27	What software, if applicable, was used to manage the data?	72
Participant checking	28	Did participants provide feedback on the findings?	N/A
Reporting			
Quotations presented	29	Were participant quotations presented to illustrate the themes/findings? Was each quotation identified? e.g. participant number	78
Data and findings consistent	30	Was there consistency between the data presented and the findings?	78
Clarity of major themes	31	Were major themes clearly presented in the findings?	76
Clarity of minor themes	32	Is there a description of diverse cases or discussion of minor themes?	76

Developed from: Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care*. 2007. Volume 19, Number 6: pp. 349 – 357

Appendix G

Project Proposal

https://osf.io/yb7xq

Appendix H

Participant Information Sheet

https://osf.io/erfqv

Appendix I

BITSEA Questionnaire

BITSEA

Parent Form

Child's name ________________________ Sex ☐ Boy ☐ Girl Date of birth ____/____/____

Parent/Guardian's name ________________________ Date of test ____/____/____

Was your child born prematurely? ☐ No ☐ Yes If yes, what was the expected date of birth? ____/____/____

Instructions: Many statements describe normal feelings and behaviors, but some describe feelings and behaviors that may be problems. Please do your best to respond to every item. Please circle the ONE response that best describes your child's behavior in the LAST MONTH.

0 = Not true / Rarely **1 = Somewhat true / Sometimes** **2 = Very true / Often**

1.	Shows pleasure when he or she succeeds (for example, claps for self).		0	1	2	*
2.	Gets hurt so often that you can't take your eyes off him or her.		0	1	2	
3.	Seems nervous, tense, or fearful.		0	1	2	
4.	Is restless and can't sit still.		0	1	2	
5.	Follows rules.		0	1	2	*
6.	Wakes up at night and needs help to fall asleep again.		0	1	2	
7.	Cries or has a tantrum until he or she is exhausted.		0	1	2	
8.	Is afraid of certain places, animals or things. *What is he or she afraid of?* ________________		0	1	2	
9.	Has less fun than other children.		0	1	2	
10.	Looks for you (or other parent) when upset.		0	1	2	*
11.	Cries or hangs onto you when you try to leave.		0	1	2	
12.	Worries a lot or is very serious.		0	1	2	
13.	Looks right at you when you say his or her name.		0	1	2	*
14.	Does not react when hurt.		0	1	2	
15.	Is affectionate with loved ones.		0	1	2	*
16.	Won't touch some objects because of how they feel.		0	1	2	
17.	Has trouble falling asleep or staying asleep.		0	1	2	
18.	Runs away in public places.		0	1	2	
19.	Plays well with other children (not including brother/sister). (Circle N if there is no contact with other children)	N	0	1	2	*
20.	Can pay attention for a long time (other than when watching TV).		0	1	2	*
21.	Has trouble adjusting to changes.		0	1	2	
22.	Tries to help when someone is hurt (for example, gives a toy).		0	1	2	*
23.	Often gets very upset.		0	1	2	
24.	Gags or chokes on food.		0	1	2	
25.	Imitates playful sounds when you ask him or her to.		0	1	2	*
26.	Refuses to eat.		0	1	2	
27.	Hits, shoves, kicks, or bites children (not including brother/sister). (Circle N if there is no contact with other children)	N	0	1	2	
28.	Is destructive. Breaks or ruins things on purpose.		0	1	2	
29.	Points to show you something far away.		0	1	2	*
30.	Hits, bites or kicks you (or other parent).		0	1	2	
31.	Hugs or feeds dolls or stuffed animals.		0	1	2	*
32.	Seems very unhappy, sad, depressed, or withdrawn.		0	1	2	
33.	Purposely tries to hurt you (or other parent).		0	1	2	
34.	When upset, gets very still, freezes, or doesn't move.		0	1	2	

The following statements describe feelings and behaviors that can be problems for young children. Some of the descriptions may be a bit hard to understand, especially if you have not seen the behavior in your child. Please do your best to respond to all statements. Please circle the ONE response that best describes your child's behavior in the LAST MONTH.

35.	Puts things in a special order over and over and gets upset if he or she is interrupted.	0	1	2
36.	Repeats the same action or phrase over and over without enjoyment. *Please give an example:* ________________	0	1	2
37.	Repeats a particular movement over and over (like rocking, spinning). *Please give an example:* ________________	0	1	2
38.	Spaces out. Is totally unaware of what's happening around him or her.	0	1	2
39.	Does not make eye contact.	0	1	2
40.	Avoids physical contact.	0	1	2
41.	Hurts self on purpose (for example, bangs his or her head). *Please describe:* ________________	0	1	2
42.	Eats or drinks things that are not edible (like paper or paint). *Please describe:* ________________	0	1	2

1 = Not at all worried **2 = A little worried** **3 = Worried** **4 = Very worried**

A.	How worried are you about your child's behavior, emotions, or relationships?	1	2	3	4
B.	How worried are you about your child's language development?	1	2	3	4

Appendix J

Interview Guide

https://osf.io/vzkdu

Appendix K

Consent Form

https://osf.io/gy9kr

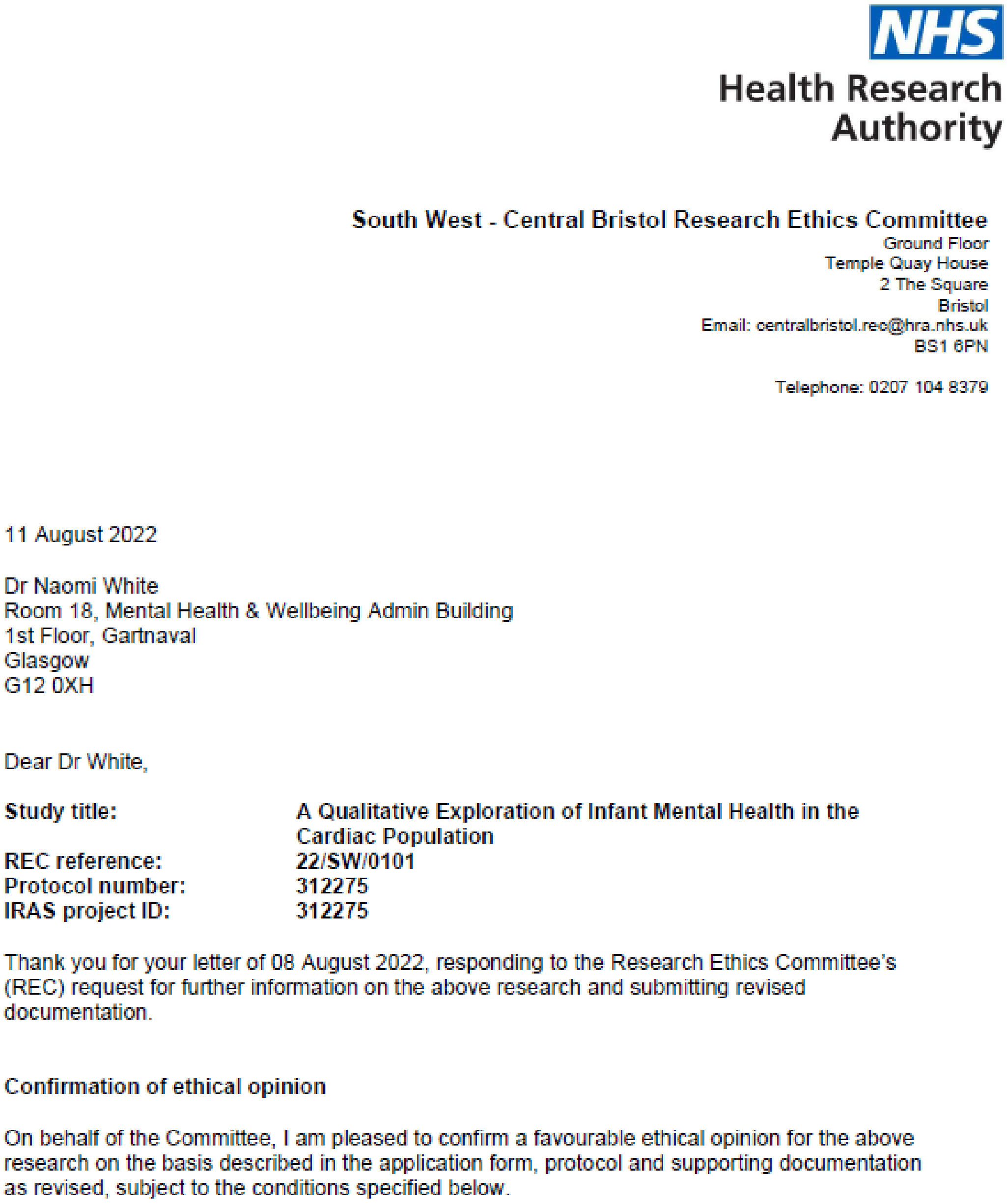
Appendix L

Coding Excerpt

Transcript	Codes
Interviewer: […] how did he cope with being in hospital?	
Participant: Like the first time round, he was only a baby, so I don't think it really had overly, much of an effect on him.	Young infants immune to early medical experiences (parents)
Interviewer: OK.	
Participant: Em… He knows now that he's older. So when we go into the hospital, he knows where we're going and as soon as we go into the room, he starts getting upset usually. Em… because, he's just had so much to happen to him. So after the second surgery he was on [type of] injections that we had to give him twice a day at home. So I did the morning and dad did night. Em… And then he had to get one or two weekly bloods as well, which is horrendous. He's got really shocking veins for getting blood out of. So it was like 2,3,4 attempts at times. And he just screamed you know, like he's a baby. Like, he doesn't know what's happening and it's sore, and why are you doing this sort of thing so. Em… But yeah, second time round he was, he was OK really. Like I don't… [pause] So before the surgery, he was fine. We were only in that night and then he was away in the morning. Em… He was really, really distressed and it's probably the most upset I've been as well, was kind of, maybe three or four days after the second surgery. Because he'd had the brain injury as well, they get sometimes, they called it cerebral irritation. And basically it's just their brain is sore, and he was crying but it was like a… a hard to explain cry, it was the worst noise I've ever heard. And that, that was the only ever time I've had to go and take a break for five minutes. [Husband] just looked at me and he was like 'Just go', so I just walked to the side for ten minutes and then I came back. Em… but it was kind of like, at that time 'Is this how he's going to be forever?'. Sorry, I always get emotional… [participant starts crying]	Older age of infant = has a MH (parents) Infants only remember at certain age (parents) Lasting impacts of medical experiences (infants) Overt distress during… appointments (infants) Threat associations with… environment (infants) Nursing within parenting Perseverance despite infants distress Disempowering experiences for infant Infants cant understand the 'why' Overt distress during… recovery from surgery (infants) Lasting memories of hospital/surgery on parents Parental anxiety Loss of child parents knew (pre-post surgery)

Appendix M

NHS REC Approval Letter



NHS
Health Research Authority

South West - Central Bristol Research Ethics Committee
Ground Floor
Temple Quay House
2 The Square
Bristol
Email: centralbristol.rec@hra.nhs.uk
BS1 6PN

Telephone: 0207 104 8379

11 August 2022

Dr Naomi White
Room 18, Mental Health & Wellbeing Admin Building
1st Floor, Gartnaval
Glasgow
G12 0XH

Dear Dr White,

Study title:	**A Qualitative Exploration of Infant Mental Health in the Cardiac Population**
REC reference:	**22/SW/0101**
Protocol number:	**312275**
IRAS project ID:	**312275**

Thank you for your letter of 08 August 2022, responding to the Research Ethics Committee's (REC) request for further information on the above research and submitting revised documentation.

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.

Good practice principles and responsibilities

The UK Policy Framework for Health and Social Care Research sets out principles of good practice in the management and conduct of health and social care research. It also outlines the responsibilities of individuals and organisations, including those related to the four elements of research transparency:

1. registering research studies
2. reporting results
3. informing participants
4. sharing study data and tissue

Conditions of the favourable opinion

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

Confirmation of Capacity and Capability (in England, Northern Ireland and Wales) or NHS management permission (in Scotland) should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise).

Guidance on applying for HRA and HCRW Approval (England and Wales)/ NHS permission for research is available in the Integrated Research Application System.

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 management permissions from host organisations

Registration of Clinical Trials

All research should be registered in a publicly accessible database and we expect all researchers, research sponsors and others to meet this fundamental best practice standard.

It is a condition of the REC favourable opinion that **all clinical trials are registered** on a publicly accessible database within six weeks of recruiting the first research participant. For this purpose, 'clinical trials' are defined as:

- clinical trial of an investigational medicinal product
- clinical investigation or other study of a medical device
- combined trial of an investigational medicinal product and an investigational medical device
- other clinical trial to study a novel intervention or randomised clinical trial to compare interventions in clinical practice.

Failure to register a clinical trial is a breach of these approval conditions, unless a deferral has been agreed by the HRA (for more information on registration and requesting a deferral see: Research registration and research project identifiers).

If you have not already included registration details in your IRAS application form you should notify the REC of the registration details as soon as possible.

Publication of Your Research Summary

We will publish your research summary for the above study on the research summaries section of our website, together with your contact details, no earlier than three months from the date of this favourable opinion letter.

Should you wish to provide a substitute contact point, make a request to defer, or require further information, please visit:
https://www.hra.nhs.uk/planning-and-improving-research/application-summaries/research-summaries/

N.B. If your study is related to COVID-19 we will aim to publish your research summary within 3 days rather than three months.

During this public health emergency, it is vital that everyone can promptly identify all relevant research related to COVID-19 that is taking place globally. If you haven't already done so, please register your study on a public registry as soon as possible and provide the REC with the registration detail, which will be posted alongside other information relating to your project. We are also asking sponsors not to request deferral of publication of research summary for any projects relating to COVID-19. In addition, to facilitate finding and extracting studies related to COVID-19 from public databases, please enter the WHO official acronym for the coronavirus disease (COVID-19) in the full title of your study. Approved COVID-19 studies can be found at: https://www.hra.nhs.uk/covid-19-research/approved-covid-19-research/

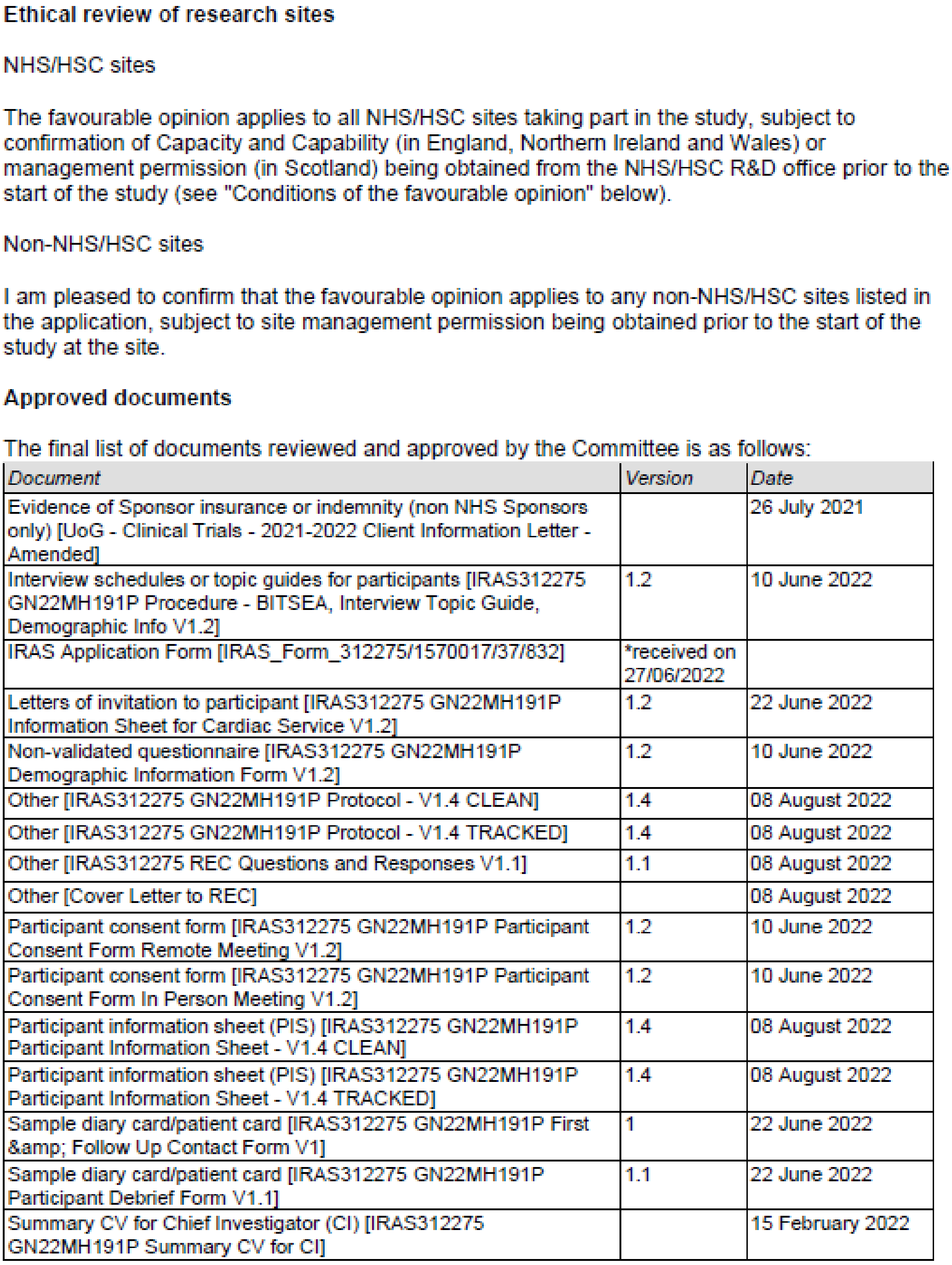
It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

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, including early termination of the study
- Final report
- Reporting results

The latest guidance on these topics can be found at https://www.hra.nhs.uk/approvals-amendments/managing-your-approval/.



Ethical review of research sites

NHS/HSC sites

The favourable opinion applies to all NHS/HSC sites taking part in the study, subject to confirmation of Capacity and Capability (in England, Northern Ireland and Wales) or management permission (in Scotland) being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Non-NHS/HSC sites

I am pleased to confirm that the favourable opinion applies to any non-NHS/HSC sites listed in the application, subject to site management permission being obtained prior to the start of the study at the site.

Approved documents

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

Document	*Version*	*Date*
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [UoG - Clinical Trials - 2021-2022 Client Information Letter - Amended]		26 July 2021
Interview schedules or topic guides for participants [IRAS312275 GN22MH191P Procedure - BITSEA, Interview Topic Guide, Demographic Info V1.2]	1.2	10 June 2022
IRAS Application Form [IRAS_Form_312275/1570017/37/832]	*received on 27/06/2022	
Letters of invitation to participant [IRAS312275 GN22MH191P Information Sheet for Cardiac Service V1.2]	1.2	22 June 2022
Non-validated questionnaire [IRAS312275 GN22MH191P Demographic Information Form V1.2]	1.2	10 June 2022
Other [IRAS312275 GN22MH191P Protocol - V1.4 CLEAN]	1.4	08 August 2022
Other [IRAS312275 GN22MH191P Protocol - V1.4 TRACKED]	1.4	08 August 2022
Other [IRAS312275 REC Questions and Responses V1.1]	1.1	08 August 2022
Other [Cover Letter to REC]		08 August 2022
Participant consent form [IRAS312275 GN22MH191P Participant Consent Form Remote Meeting V1.2]	1.2	10 June 2022
Participant consent form [IRAS312275 GN22MH191P Participant Consent Form In Person Meeting V1.2]	1.2	10 June 2022
Participant information sheet (PIS) [IRAS312275 GN22MH191P Participant Information Sheet - V1.4 CLEAN]	1.4	08 August 2022
Participant information sheet (PIS) [IRAS312275 GN22MH191P Participant Information Sheet - V1.4 TRACKED]	1.4	08 August 2022
Sample diary card/patient card [IRAS312275 GN22MH191P First & Follow Up Contact Form V1]	1	22 June 2022
Sample diary card/patient card [IRAS312275 GN22MH191P Participant Debrief Form V1.1]	1.1	22 June 2022
Summary CV for Chief Investigator (CI) [IRAS312275 GN22MH191P Summary CV for CI]		15 February 2022

Summary CV for student [IRAS312275 GN22MH191P CV of Student/Lead Researcher]		20 May 2022
Summary CV for student [NHS GGC Lone Working Policy]	3	01 December 2021
Summary CV for student [UofG Lone Working Policy - Updated version]		10 May 2022
Validated questionnaire [IRAS312275 GN22MH191P BITSEA Parent Questionnaire]	1	

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.

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 Learning

We are pleased to welcome researchers and research staff to our HRA Learning Events and online learning opportunities– see details at:
https://www.hra.nhs.uk/planning-and-improving-research/learning/

IRAS project ID: 312275 Please quote this number on all correspondence

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

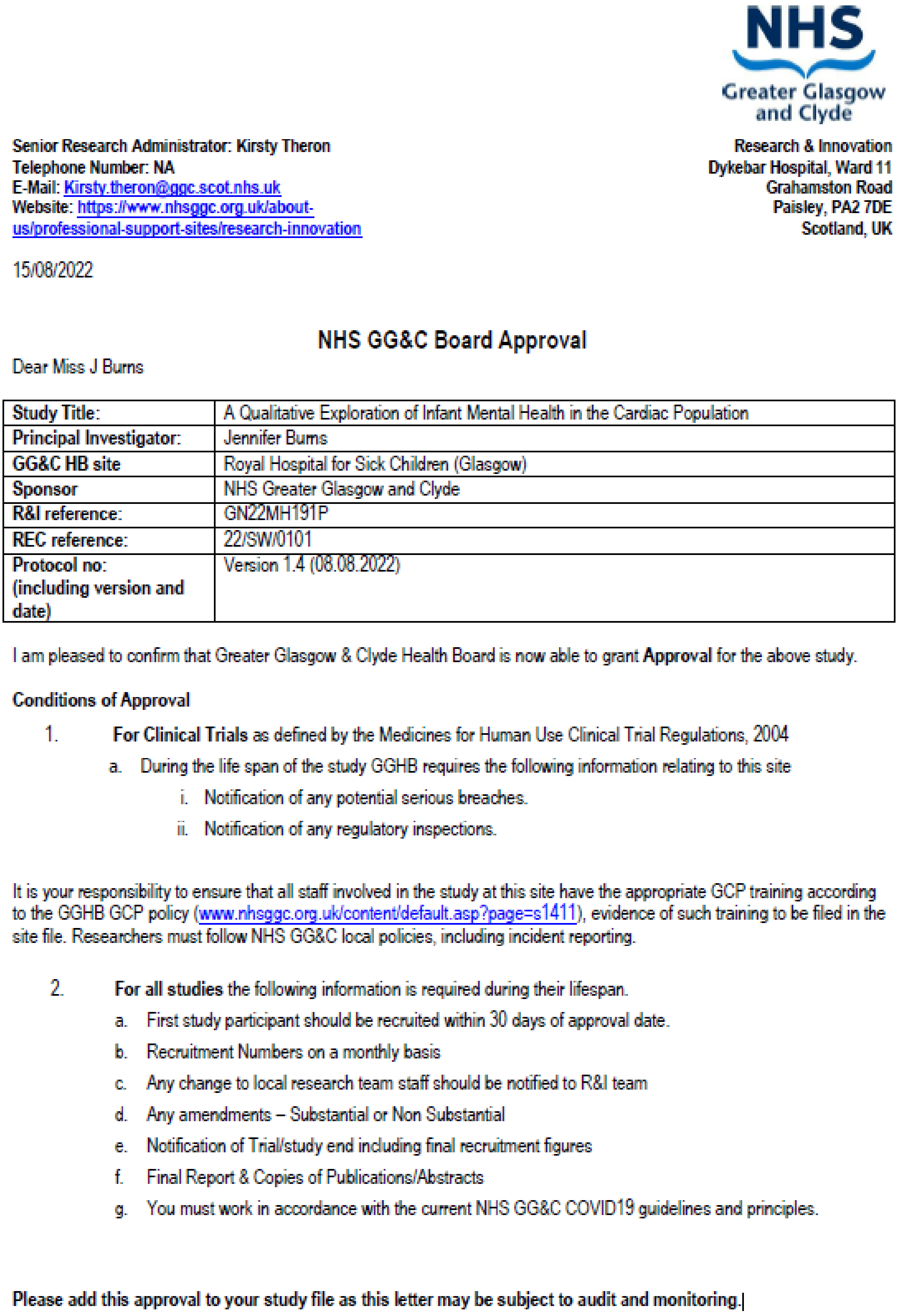
Yours sincerely

Dr Julie Woodley
Chair

Email:centralbristol.rec@hra.nhs.uk

Appendix N

NHS GGC R&I Approval Letter



Senior Research Administrator: Kirsty Theron
Telephone Number: NA
E-Mail: Kirsty.theron@ggc.scot.nhs.uk
Website: https://www.nhsggc.org.uk/about-us/professional-support-sites/research-innovation

Research & Innovation
Dykebar Hospital, Ward 11
Grahamston Road
Paisley, PA2 7DE
Scotland, UK

15/08/2022

NHS GG&C Board Approval

Dear Miss J Burns

Study Title:	A Qualitative Exploration of Infant Mental Health in the Cardiac Population
Principal Investigator:	Jennifer Burns
GG&C HB site	Royal Hospital for Sick Children (Glasgow)
Sponsor	NHS Greater Glasgow and Clyde
R&I reference:	GN22MH191P
REC reference:	22/SW/0101
Protocol no: (including version and date)	Version 1.4 (08.08.2022)

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. Researchers must follow NHS GG&C local policies, including incident reporting.

2. **For all studies** the following information is required during their lifespan.
   a. First study participant should be recruited within 30 days of approval date.
   b. Recruitment Numbers on a monthly basis
   c. Any change to local research team staff should be notified to R&I team
   d. Any amendments – Substantial or Non Substantial
   e. Notification of Trial/study end including final recruitment figures
   f. Final Report & Copies of Publications/Abstracts
   g. You must work in accordance with the current NHS GG&C COVID19 guidelines and principles.

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,

Kirsty Theron
Senior Research Administrator

CC: Dr N White, Dr CM Sardar

Appendix O

Participant Demographic Details

Caregiver Characteristics		Responses (*n* = 7)	
		n	%
Relation to infant			
	Mother	7	100%
	Father	0	-
	Other	0	-
Ethnicity			
	White	7	100%
	Black or African American	0	-
	Asian	0	-
	Mixed or Multiple Ethnicities	0	-
	Other	0	-
Caregiver Age			
	18-20 years	0	-
	21-29 years	1	14.3%
	30-39 years	4	57.1%
	40-49 years	2	28.6%
	50 years or over	0	-
Total Number of Children			
	1	5	71.4%
	2	2	28.6%
	3 or more	0	-
Caregiver Employment Status			
	Employed full time	1	14.3%
	Employed part time	3	42.8%
	Self employed	2	28.6%
	Not working	1	14.3%
Marital Status			
	Single	0	-
	Cohabiting	1	14.3%
	Married	6	85.7%
	Divorced	0	-
	Other	0	-
Scottish Index of Multiple Deprivation			
	Not specified	1	14.3%

Quintile 1 (Most Deprived)	1	14.3%
Quintile 2	1	14.3%
Quintile 3	2	28.6%
Quintile 4	2	28.6%
Quintile 5 (Least Deprived)	0	-

Infant Characteristics	Responses (*n* = 7)	
	n	*%*
Infant Age		
12-18 months	2	28.6%
19-24 months	3	42.8%
25-32 months	2	28.6%
33-36 months	0	-
Infant Gender		
Male	3	42.9%
Female	4	57.1%
Other	0	-
Number of Cardiac Surgeries to Date		
1	3	42.8%
2	2	28.6%
3 or more	2	28.6%
Total Time Spent in Hospital in Relation to Diagnosis		
0-7 weeks	3	42.9%
2-6 months	4	57.1%
7-11 months	0	-
1 year or more	0	-
Previous Support from link Clinical Psychologist		
Yes	3	42.9%
No	4	57.1%
BITSEA		
Possible Social-Emotional Problem Range		
Yes	3	42.9%
No	4	57.1%
Possible Development Delay Range		
Yes	1	14.3%
No	6	85.7%

Appendix P

Descriptive Narratives for Themes 4 & 5

In terms of promoters of IMH, caregivers recounted many instances of nursing staff being attuned and sensitive to their infant's needs, out with their physical health condition. This included nursing staff recognising and tailoring either their interaction style or the environment to suit infants' preferences, at times even before the caregivers had identified these themselves. Implicit within this was a sense of valuing health care professionals engaging with the infant as an infant, as opposed to a patient. Nurses also acted as substitute caregivers for the parents at times, particularly during the lockdown restrictions as parents were unable to access their own support networks. This support from nursing staff to parents, in turn, appeared to enhance caregivers' capacity to care for their infant's needs. All caregivers also identified the benefits of health care professionals engaging in some level of interaction or play with the infant during hospitalisation, either for stimulation, distraction, or to help them regulate after a difficult medical experience. Play was viewed by caregivers as a way for infants to develop trust with the health care professionals, which was imperative given the potentially traumatic nature of some other necessary interactions between them. Even brief moments of playfulness from professionals towards the infant appeared to encourage the infants' engagement and co-operation in medical interventions thereafter. As Sara described: *"Any nurse who gave time to her just to play was definitely an advantage. Like, because she is quite nervous of people"*. Many caregivers found that their infants were calmer and more accepting of medical interventions if they thought their caregivers were assisting with this in some way. For example, if caregivers were encouraged by medical staff to hold the infant during interventions and/or the equipment, where possible, infants were more co-operative.

Destabilisers to IMH during hospitalisation included aspects of the environment, such as the physical space not always being tailored to infants' needs, and the lack of privacy or autonomy infants had regarding their physical space. Caregivers identified practicalities, such as not having access to a highchair or appropriately sized clothing, as detrimental, and this evoked a sense of infants not feeling held in mind by hospital systems. Moreover, as this cohort of infants experienced hospitalisation and surgery during the COVID-19 pandemic, there was a heightened sense of loss in terms of limited socialisation for the infant with same-aged peers and, or extended family. Caregivers described a sense of entrapment and confinement to their hospital rooms, exacerbated further because of the COVID-19 restrictions. In some instances, this seemed to contribute to enmeshment between the caregivers and infants and may to some extent relate to the aforementioned relational changes in infants post-hospitalisation. As most infants were non-verbal at the time of hospitalisation and surgery, caregivers perceived that health care professionals had difficulties gauging their internal states, including their pain levels; this meant pain-relief could not always be tailored appropriately to the infants' thresholds, which led to increased distress for the infant. Various sensory aspects of certain medical interventions were also thought to be particularly distressing for the infants, including the sensation of the blood pressure machine or the gel used within echocardiograms. Caregivers recognised the challenges of not being able to pre-warn infants of these upcoming sensations or communicate with them regarding the need for these. Common across many accounts were recollections of health care professionals persevering with medical interventions despite high levels of infant distress, which evoked a sense of infants being disempowered and highly threatened. As Beth described when discussing her infant being given medication*: "It was kind of, pin him down, which was horrific"*. Louise similarly reported: *"He had an NG tube and they were forcing it obviously down, so that was, that was the start".* Moreover, a small number of participants described instances where it had either been suggested by staff that they leave prior to a difficult medical intervention, or they were unable to stay overnight due to departmental policies/restrictions, meaning the infant had had to self-soothe at high moments of distress, when they would have otherwise accessed their caregiver.