

**Stigma, Concealment, Illness Perceptions and
Psychosocial Difficulties in Children with
Physical Health Conditions and their Parents**

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Thesis declaration form

I confirm that the work presented in this thesis is my own. Where information has been derived from other sources, I confirm that this has been indicated in the thesis.

Signature:



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Date: 18th June 2020

Overview

This thesis sets out to better understand the stigma experiences of children with physical health conditions, alongside factors of illness perceptions, concealment and psychosocial difficulties. Data collection for the empirical study was conducted jointly with Jemma Ambrose.

Part one is a systematic literature review, which sought to better understand illness perceptions and stigma experiences in children with chronic health conditions, and the inter-relationship between these two constructs. A thematic synthesis of qualitative studies investigating stigma and illness perceptions is presented, with the resultant themes and sub-themes which were identified.

Part two is an empirical paper, of an exploratory quantitative study which sought to investigate the relationships between stigma, concealment, illness perceptions and psychosocial difficulties in children with physical health problems and their parents. Both child and parent stigma were found to be independently associated with children's psychosocial difficulties, as well as with the concealment of the child's health condition by children and their parents. No child and parent-rated factors were associated with each other. The implication of these findings and avenues for future research are discussed.

Part three is a critical appraisal of the research process documented in the empirical paper. Issues which emerged in the development and conducting of the research are discussed, along with personal reflections on the research process.

Impact Statement

This thesis informs research and clinical practice related to stigma and related experiences in children with physical health conditions. The first part is a systematic literature review focussed on stigma and illness perceptions in children with chronic health conditions, and provides a thematic synthesis of qualitative studies. The results of this review highlighted different elements of the stigma experiences of children with chronic health conditions, and provide a rationale for investigating internalised stigma and perceived stigma separately in future research. Medication and symptom management emerged as an element of how children's illness perceptions, a theme which is currently lacking from quantitative research studies. Finally, areas of intersection between stigma and illness perceptions provide possible targets for interventions.

The second part of this thesis documents an empirical study which investigated the relationships between physical health stigma, concealment, illness perceptions and psychosocial difficulties in children with physical health problems and their parents. Child and parental stigma were both independently associated with children's psychosocial difficulties in this study: this highlights the importance of professionals having an awareness of stigma, and for them to routinely ask children with physical health conditions and their parents about their stigma experiences. Given that child and parental factors were not associated with each other, both perspectives should be seen as providing useful insight into psychosocial difficulties, even if their accounts appear incongruent with each other. Finally, concealment was associated with stigma but not with children's psychosocial difficulties, with the implication that it should not be assumed that professionals should discourage concealment, but rather support families in making disclosure decisions. The association of stigma with illness perceptions and concealment also opens up avenues for future research around interventions to support children with physical health conditions.

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Part 1 – Systematic Literature Review

Stigma Experiences and Illness Perceptions in Children with Chronic Health Conditions: A Thematic Synthesis

1. Abstract

Introduction: Stigma and illness perceptions are related to psychological distress in children with chronic health conditions. Despite children with chronic health conditions being at greater risk of experiencing mental health problems, this area of the literature is still in development. A better understanding of stigma and illness perception could have clinical implications on interventions offered, and lay the foundation for future research.

Aim: To better understand illness perceptions and stigma experiences in children with chronic health conditions, and the inter-relationship between these constructs.

Method: A thematic synthesis of qualitative research examining stigma and illness perceptions from the perspective of children with chronic health conditions was conducted. The quality of studies was appraised using the Critical Appraisal Skills Programme (CASP) checklist.

Results: Seventeen studies were included in the review. Two analytic themes of: 'components of illness perceptions' and 'components of stigma experiences', and eight sub-themes were generated.

Conclusion: The findings aligned with previous research of children with chronic health conditions and with the wider stigma literature, but also highlighted the importance of considering the component parts of stigma and illness interventions. Future research investigating these components and how they relate to each other could be used to guide psychosocial interventions.

2. Introduction

The prevalence of chronic health conditions in children and adolescents is estimated to be between 13-27% and appears to be increasing (Van Cleave et al., 2010). Children with chronic health conditions are also at a greater risk of experiencing psychological distress (Hysing et al., 2007) or developing mental health problems (Ferro et al., 2015). However, the literature around the psychosocial factors influencing the experiences of living as a child with a chronic health condition is in its infancy. A greater understanding of the factors contributing to the physical and mental health experiences of children with chronic health conditions could inform the care they are offered.

2.1. Stigma

Stigma is associated with individuals who have attributes which deviate from dominant societal norms, in a way that is seen to 'discredit' or 'taint' them (Goffman, 1963). Stigma is therefore an inherently social phenomenon, with societal responses determining which attributes are stigmatised. The assumptions and behaviours which arise based on an individual's stigmatised attributes can be classified as follows: a *stereotype* is defined as a negative belief against a group; *prejudice* involves the agreement with such beliefs; and *discrimination* is understood as the behavioural response to such negative beliefs (Corrigan et al., 2002). Although stigma can manifest in how the general population views and responds to stigmatised individuals in what has been termed *public stigma* (Bos et al., 2013), these attitudes can also be internalised by those with stigmatised identities in what has been termed *self-stigma* (Bos et al., 2013). The negative consequences of stigma include lower self-esteem (Corrigan et al., 2006), less treatment seeking (Jennings et al., 2015) and increased psychological distress (Quinn et al., 2009).

There is a growing recognition of the stigma experienced by children with chronic health conditions, with one study finding that half of children with epilepsy rated social stigmatisation as the worst part of having epilepsy (Vanstraten et al., 2012). Children with chronic health conditions report experiences of peer rejection, including bullying, being stared at or being excluded (Elliott et al., 2005; McMurray et al., 2001; Strauss et al., 2007). As children with chronic health conditions progress into middle childhood and adolescence, this area of 'difference' coincides with a developmental stage when peer belonging is perceived to be increasingly important (Pittman et al., 2007). Peer rejection has also been found to moderate the association between pain and depressive symptoms in children with juvenile rheumatic disease (Sandstrom et al., 2004), suggesting that stigma experiences may have a significant contribution to the psychological, social and physiological experiences of in children with chronic illnesses.

As in the broader stigma literature, there is evidence of stigma having adverse consequences on the psychological wellbeing of children with chronic health conditions. In children with epilepsy, stigma has been associated with depressive symptoms, worry and low self-esteem (Austin et al., 2004; MacLeod et al., 2003). There is similar evidence linking stigma with depressive symptoms in inflammatory bowel disease (Gamwell et al., 2018) and with lower health-related quality of life in children with facial differences (Masnari et al., 2013a). The increased risk of mental health problems in children with chronic health conditions means that they may relate to several stigmatised identities, but the focus on this review is on physical health stigma.

2.2. Illness Perceptions

Whereas stigma is associated with 'difference' from others, the way in which children perceive their chronic health condition is also an important aspect in understanding their illness-related experiences. Illness perceptions are understood to encapsulate

how children with chronic conditions think about and understand their condition and the resulting functional limitations (Ramsey et al., 2016), and whether they see these as positive or negative. Illness perceptions are important because they can predict the types of coping behaviours children engage in, which may be considered as either 'adaptive' or 'maladaptive' by parents and professionals (Austin et al., 1991). When considering illness perceptions, it is worth noting that there has been criticism of how well this construct translates across cross-cultural experiences and the measurement of this across different cultures and languages (Brzoska et al., 2010), and that the majority of the current literature is based on Western populations. However, within the available literature negative illness attitudes have been associated with depressive symptoms, stress and frequency of emotions about the health condition (Austin et al., 2006; le Coq et al., 2000; Wagner et al., 2008), whereas positive illness attitudes have been associated with lower levels of mental health symptoms (LeBovidge et al., 2005) and better health outcomes in children with chronic conditions (Murphy, 1974). Therefore, despite some of the limitations of this construct, a better understanding of children's illness perceptions could inform how to target psychological interventions, and could be another key factor in understanding illness experiences.

2.3. Stigma and Illness Perceptions

Whereas illness perceptions involve children's individual attitudes or relationship to their conditions, stigma is a social construct related to 'difference' from others. Taken together, these constructs therefore represent a potential interplay between how children themselves see their chronic health condition and how they believe others perceive them. A preliminary search of the literature on stigma and illness perception revealed a small cluster of quantitative studies investigating these concepts in epilepsy. These studies found an association between self-stigma and illness perceptions in children with epilepsy (Austin et al., 2004; Funderburk et al., 2007), with higher levels of stigma being associated with more negative illness perceptions.

Understanding this relationship has clinical implications, because it could guide future research about whether clinical interventions focussed on one of these areas could affect the other.

However, the quantitative literature was sparse and had a number of limitations. Most studies used two components of a generic health-related quality of life measure to measure stigma and illness perceptions (Devinsky et al., 1999; Stevanovic, 2007; Zamani et al., 2014), which provides limited insight into how they are experienced by children with chronic health conditions. Specific measures of child physical health stigma (Child Stigma Scale) and illness perceptions (Child Attitude towards Illness Scale; CATIS) have been developed for children with epilepsy (Austin et al., 1993; Austin et al., 2004). However, a recent systematic review of the CATIS only identified one study which considered illness attitudes alongside stigma (Ramsey et al., 2016). Furthermore, although there are some similarities in how the CATIS operationalises illness perceptions compared to adult models such as the common sense model of illness perceptions (Diefenbach et al., 1996), the measure was developed primarily based on the experiences of children with epilepsy, and may therefore not encompass the wider experiences of children with chronic health conditions. The Child Stigma Scale was similarly developed for children with epilepsy, and the vast majority of the quantitative literature is focussed on this population. The qualitative literature includes a broader range of chronic health conditions, and also provides a richer insight into how children relate to their illness and experience difference. In this review, the focus will be on the child's perspectives of stigma and illness perceptions (as opposed to caregivers or professionals), due to illness perceptions being inherently related to the child's way of relating to their condition.

2.4. Aims and Rationale

This review aims to provide a richer understanding of stigma experiences and illness perceptions in children across all chronic health conditions, as well as how these two

constructs overlap. Qualitative research will be considered in this review, due to the sparsity of the quantitative literature, the predominance of epilepsy-based research and limitations in how stigma and illness perceptions have been measured. By considering richer descriptions of these constructs and how they apply across a range of chronic conditions, this review aims to provide a useful foundation for future research.

To the author's knowledge, there are no reviews which have specifically sought to examine stigma and illness perceptions across all chronic health conditions, although previous reviews of the general experiences of children with chronic health conditions have highlighted findings related to these constructs (e.g. Chong et al., 2016; Lambert et al., 2015; Tong et al., 2012).

This review had the following aims:

1. To better understand physical health stigma experiences and illness perceptions in children with chronic health conditions.
2. To better understand the inter-relationship between these two constructs.

3. Method

3.1. Search strategy

Searches were conducted on the following electronic databases in July 2019 to identify eligible papers for the inclusion in this review: MEDLINE (1946–present), PsychINFO (1806–present), Embase (1974–present), CINAHL (1965–present) and Web of Science (1900–present). No limitations were set based on the year of publication. Search terms were focussed on three areas: children with chronic health conditions, stigma and illness perceptions (see Table 1). The search terms of systematic reviews which also investigated chronic illnesses were used to inform

Table 1

Search Terms and Strategy

Key search terms	Search Strategy
Child	child* OR young person* OR teen* OR youth* OR youngster* OR adolescent* OR kid* OR paediatric* OR pediatric*
Chronic illness	physical illness OR physical disease OR chronic illness OR chronic disease OR long term conditions OR long term condition OR arthritis OR asthma OR cancer OR chronic fatigue syndrome OR cleft OR cystic fibrosis OR deaf OR diabetes OR epilepsy OR headache OR heart disease OR hearing impairment OR inflammatory bowel disease OR kidney disease OR liver disease OR migraine OR rheumatism OR sickle cell OR spina bifida OR visual impairment OR respiratory OR dermat* OR chronic pain
Stigma	stig*
Illness perception	attitud* OR perception OR illness representations OR illness beliefs

which conditions were included in the search (e.g. Pinguart et al., 2011). Search terms within each area were combined with the Boolean term 'OR', and between each area combined with 'AND'. No limiters were applied. Ancestry searching was also conducted, by manually searching the bibliographies of potentially eligible papers and relevant review papers which emerged from the search.

Studies were deemed eligible for inclusion if participants were aged 0-18 years, had any chronic health condition, and if both stigma and illness perceptions were examined either explicitly or emerged as incidental themes or sub-themes in the data analysis. Papers referring to 'difference', deviations from 'normal' or experiences of discrimination related to chronic health conditions were included as encompassing stigma experiences, even if the word 'stigma' was not used in the theme. Inclusion for illness perceptions involved participants expressing attitudes, beliefs or emotional responses related to their condition. Only qualitative studies were included, and both quantitative and mixed-method studies were excluded. Non-English language, review

papers and non-peer reviewed studies such as conference abstracts, commentaries, dissertations and book chapters were excluded. To maintain a focus on children's perspectives, studies were also excluded if they included the perspectives of those other than the child with a chronic health condition, such as family members, teachers or healthcare professionals.

3.2. Study Selection

The study selection process was conducted in two stages. In the first stage, all titles and abstracts were screened and duplicates were removed. In the second stage, the full texts of all potentially eligible papers were read in full and the study selection criteria were used to determine eligibility. Reasons for exclusion were documented at each stage.

3.3. Quality Assessment

The Critical Appraisal Skills Programme (CASP, 1998) was used to appraise the studies included in this review. This is a 10-item checklist is used to critically evaluate qualitative studies using the following criteria: clarity of research aims; appropriateness of methodology; appropriateness of research design; appropriateness of recruitment strategy; data collection; adequate consideration of the relationship between researchers and participants; ethical considerations; rigorousness of data analysis; clarity of findings; and how valuable the research is. For each item, the reviewer is required to provide a rating of "Yes", "No" or "Can't tell". In this review, the author rated nineteen studies using the CASP framework. A second member of the research team independently reviewed ten of the studies, and inter-rater reliability analysis using the Kappa statistic was performed to determine consistency among raters.

3.4. Data analysis and synthesis

A three-stage thematic synthesis approach as described by Thomas et al. (2008) was used to analyse the data from eligible studies. This method was adopted because it goes beyond a thematic analysis of findings by generating concepts to answer the review question, whilst differing from other common qualitative meta-analysis methods like a meta-ethnography which aim to develop theories or models. In this process, all eligible papers were included regardless of the method of data collection, analysis or epistemological position. Although this is consistent with a thematic synthesis approach, other qualitative synthesis methodologies may focus on particular study designs due to concerns about the validity of combining different approaches (Soilemezi et al., 2018).

First, inductive line-by-line coding was used to analyse all data relating to stigma or illness perceptions, including lists of themes, quotes, and descriptive text of relevant themes. After the initial coding, all data was re-read and axial coding was used to identify any additional levels of coding. This also allowed for what Thomas et al. (2008) described as the 'translation' of concepts between studies. Secondly, descriptive themes were identified by grouping codes and constructing these within a hierarchical tree structure. Finally, analytical themes were generated from the descriptive themes, by 'going beyond' the original findings to answer the review questions based on inferences of the researcher. This is similar to the process of 'third-order interpretations' carried out in meta-ethnography methodology. In the context of this study, the process of deriving analytic themes from the descriptive themes was influenced by the research questions about stigma and illness perceptions. Once initial themes were generated by the first author, a member of the research team reviewed these findings and final themes were agreed.

4. Results

An adapted PRISMA (Moher et al., 2009) flow diagram in Figure 1 depicts the stages of identification, screening and establishing eligibility of the final included studies. Of the 1,995 studies which were identified by the initial search, 766 were excluded due to being duplicates and 1191 papers were excluded as a result of not meeting the selection criteria. In addition to the remaining 38 potentially eligible papers, 12 papers were identified by a hand search of bibliographies of potentially eligible papers and of review papers which emerged in the search. Fifty full-text articles were assessed for eligibility, of which 33 were excluded for reasons presented in Figure 1. A total of 17 studies were included, and Table 2 presents a summary of these studies.

4.1. Description of studies

Six studies were conducted in the United States (Christian et al., 1997; Herrman, 2006; Salazar et al., 2014; Velsor-Friedrich et al., 2004; Walker et al., 2014), three in the United Kingdom (McEwan et al., 2004; McMurray et al., 2001; Moffat et al., 2009), three in Canada (Elliott et al., 2005; Moola et al., 2011; Protudjer et al., 2009), two in Taiwan (Chen et al., 2010; Wang et al., 2010), one in Norway (Winger et al., 2014), one in Ireland (Benson et al., 2015a), and one in Palestine (Nahal et al., 2019). In total, 368 participants took part in all the studies.

A range of chronic health conditions were included: five studies focussed on young people with epilepsy (Benson et al., 2015a; Chen et al., 2010; Elliott et al., 2005; McEwan et al., 2004; Moffat et al., 2009), three on young people with Type I diabetes (Freeborn et al., 2013; Herrman, 2006; Wang et al., 2010), three on young people with asthma (Protudjer et al., 2009; Velsor-Friedrich et al., 2004; Walker et al., 2014), two on young people with congenital heart disease (McMurray et al., 2001; Moola et al., 2011), one on young people with cystic fibrosis (Christian et al., 1997), one on young people with spina bifida (Nahal et al., 2019), one on young people with

Figure 1

PRISMA diagram representing the identification and selection process

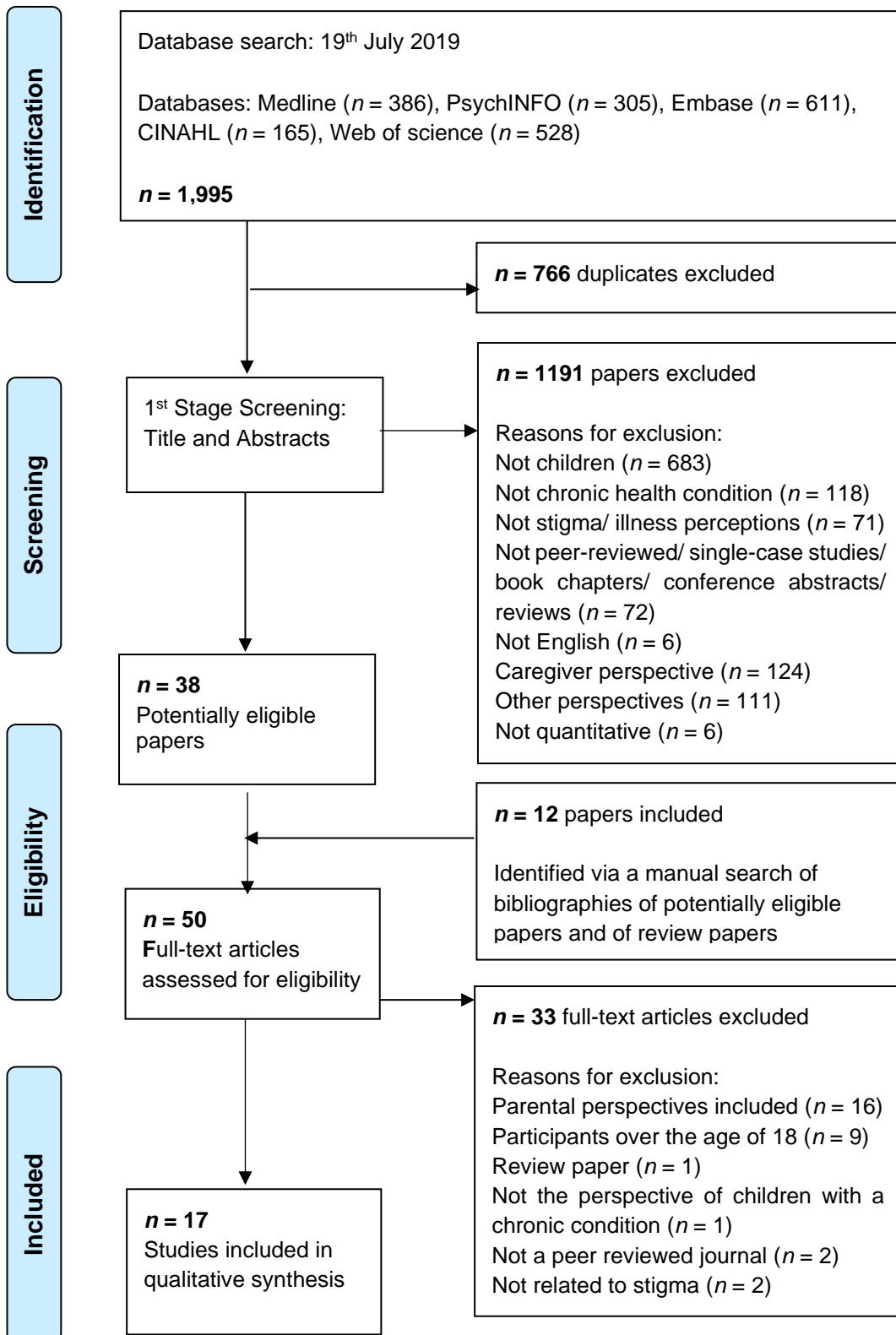


Table 2

Summary of Findings

Authors (year), Country	Chronic Health Condition	Aim/ Objective	Sample	Method	Analysis	Key Findings
Benson et al. (2015a) Ireland	Epilepsy	To identify contextual factors presenting as obstacles to disclosing children's diagnosis of epilepsy	n = 29 young people with epilepsy Age: 6-16 years Gender: 17 female, 12 male Ethnicity: Not stated	Semi-structured interviews, using a topic guide	Thematic analysis	Five core themes, with sub-themes: 1. Desire for normalcy Subthemes: <i>Feelings of differentness; Minimizing different treatment</i> 2. Out of sight but in the mind Subthemes: <i>Invisibility of epilepsy; Epilepsy and the brain</i> 3. Contending with negative responses to disclosure Subthemes: <i>Anticipating negative responses; Actual negative responses</i> 4. The complexity of epilepsy Subthemes: <i>Difficult to explain to others; Challenging for peers to understand</i> 5. Self and others' perceptions of epilepsy Subthemes: <i>Epilepsy as something private; Epilepsy as something negative; Others' perceptions</i>

Chen et al. (2010)	Epilepsy	To explore the experiences of children in Taiwan who are living with epilepsy	n = 15 young people with epilepsy Age: 7-12 years Gender: 10 female, 5 male Ethnicity: Not stated	Semi-structured interviews, using an interview guide	Phenomenological method	Two main themes, with sub-themes and categories 1. Living with epilepsy Subthemes: <i>Illness-related experiences</i> <i>School-related issues</i> 6. Coping with epilepsy Subthemes: <i>Strategies to manage seizures</i> ; <i>Seeking support from family</i>
Taiwan						
Christian et al. (1997)	Cystic fibrosis	To explore conceptualisations of growing up with cystic fibrosis	n = 20 young people cystic fibrosis Age: 12-18 years Gender: 12 female, 8 male Ethnicity: 17 'White', 3 'Native American'	Semi-structured interviews	Grounded theory and constant comparative method (Glaser et al., 1968)	One central phenomenon: Reducing a sense of difference Protective strategies to reduce sense of difference: 1. Keeping secrets 2. Hiding visible differences 3. Discovering a new baseline
United States						
Elliott et al. (2005)	Epilepsy	To explore how young people perceive the impact of epilepsy on their quality of life	n = 49 young people with epilepsy Age: 7-18 years	Semi-structured interviews, using a topic guide	Grounded theory	Four main themes, with an overarching theme of seizures as a barrier to normalcy . 1. Physical domain
Canada						

			<p>Gender: 25 female, 24 male</p> <p>Ethnicity: 46 'Caucasian', 2 'Asian-Canadian' and 1 'African-Canadian'</p>			<p>Subthemes: <i>Fatigue; Need for more sleep; Anergia, inertia</i></p> <ol style="list-style-type: none"> Emotional/behavioural domain Subthemes: <i>Unpredictability of seizures and loss of control; Intermittent feelings of sadness, depression; Frustration and anger</i> Social domain Subthemes: <i>Variability of meaning of close friendships; Barriers to inclusion: personal; Barriers to inclusion: peers; Barriers to inclusion: limits imposed by parents and others; Resilience: taking control</i> Cognitive/academic domain Subthemes: <i>Fixed or ongoing deficits; Intermittent transient disconnections; Impaired attention or concentration</i>
Freeborn et al. (2013) United States	Diabetes Mellitus (Type I)	To identify challenges experienced by young people with Type I diabetes from their own perspective	<p>n = 16 young people with Type I diabetes</p> <p>Age: 6-18 years</p> <p>Gender: 5 female, 11 male</p> <p>Ethnicity: 16 'Caucasian'</p>	Focus groups, using open-ended questions	Inductive analysis, with open, axial and selective coding procedures used	<p>Three themes related to challenges of doing with Type I Diabetes:</p> <ol style="list-style-type: none"> Low blood glucose Self-care activities Feeling different and/or alone

Herrman (2006) United States	Diabetes Mellitus (Type I)	To explore children's beliefs about the costs and rewards of diabetes and diabetes treatment	<i>n</i> = 17 young people with diabetes Age: 8-15 years Gender: 10 female, 7 male Ethnicity: Not stated	Semi-structured interviews, using an interview guide	Social exchange theory informed data analysis (Ethnographic informed approach)	Eight categories : 1. Costs of diabetes mellitus 2. Rewards of diabetes mellitus 3. Costs of treatment 4. Rewards of treatment 5. Costs to family 6. Rewards to family 7. Ways to manage costs 8. Ways to increase rewards
McEwan et al. (2004) United Kingdom	Epilepsy	To describe epilepsy experiences, contribute to understanding of quality of life (QoL), and explore issues of transitions to adulthood	<i>n</i> = 22 young people with epilepsy Age: 12-18 years Gender: 16 female, 6 male Ethnicity: Not stated	Focus groups, including written information from participants	Thematic coding	Two main themes, with subthemes 1. Adolescent Development (Identity formation) <i>Subthemes: Peer Acceptance; Development of Autonomy; School-Related Issues; Future; Epilepsy As Part of Me</i> 2. Epilepsy related variables <i>Subthemes: Medication Issues; Seizures; Knowledge of Epilepsy; Sense of Uncertainty</i>
McMurray et al. (2001) United Kingdom	Congenital heart disease	To examine the experiences of children and adolescents living with congenital heart disease	<i>n</i> = 37 young people with congenital heart disease Age: 11-18 years	Interviews, using a topic guide	Framework approach (Ritchie et al., 2002)	Five over-arching themes: 1. Coping with the presence of disease 2. Limited by impairment 3. Exclusion by others 4. Bullying and discrimination 5. Life improvement

			Gender: 17 female, 20 male			
			Ethnicity: Not stated			
Moffat et al. (2009)	Epilepsy	To investigate the impact of epilepsy on children's quality of life, and to identify concerns related to the age of children,	n = 22 young people with epilepsy Age: 7-12 years Gender: 11 female, 11 male Ethnicity: Not stated	5 Focus groups and 2 semi-structured interviews	Grounded theory	Two major themes, with sub-themes 1. Things to do with growing up Subthemes: <i>Social impact; Peer Acceptance; School-related issues; Development of autonomy; Epilepsy and self</i> 2. Things to do with your epilepsy Subthemes: <i>Antiepileptic medication; Seizures; The future; Medical experiences</i>
United Kingdom						
Moola et al. (2011)	Congenital heart disease	To explore social barriers to engaging in physical activity for young people with congenital heart disease	n = 17 young people with congenital heart disease Age: 11-17 years Gender: 10 female, 7 male	Multi-modal: semi-structured interviews and a "draw-and-write technique" artwork task	Grounded theory	Four themes: 1. What I Wish You Knew 2. Secret Keeping: Negotiating Disclosure in Health and Physical Activity 3. The Things That Stand in My Way: Barriers Encountered During Physical Education 4. The Normal/ Abnormal CHD Body: Contesting the Normal Body in Health and Physical Activity
Canada						

			Ethnicity: Not stated			
Nahal et al. (2019) Palestine	Spina bifida	To explore the lived experiences of children with spina bifida in the West Bank area of Palestine	n = 10 young people with spina bifida Age: 7-18 years Gender: 4 female, 6 male Ethnicity: Not stated	Semi-structured interviews, using an interview guide	Phenomenological hermeneutical approach	Three themes, with sub-themes 1. Experiencing negative self-concept Subthemes: <i>Resentment against disability; Powerlessness and dependency; Struggle with being different</i> 2. Experiencing vulnerability Subthemes: <i>Living with stigmatisation; Living with limitations: Risk to the body and self</i> 3. Obtaining a sense of security Subthemes: <i>Belonging with the family; Belonging with peers</i>
Protudjer et al. (2009) Canada	Asthma	To better understand the strategies used by children with asthma to normalise their lives	n = 22 young people with asthma Age: 11 years Gender: 11 female, 11 male Ethnicity: White Canadian (16/20)	Semi-structured interview, using an interview guide	Thematic analysis	Six overall themes: 1. Acknowledging the impact of asthma 2. Minimising the health impact of asthma 3. Stressing normality 4. Emphasising abilities 5. Adaptation in daily living 6. Managing asthma symptoms with medications

Salazar et al. (2014) United States	Irritable bowel disease (IBD)	To investigate the knowledge of children with IBD and their perceptions of an IBD summer camp	<i>n</i> = 25 young people with IBD (2 groups: attended camp/ did not attend camp) Age: 8-7 years Gender: 16 female, 9 male Ethnicity: Not stated	Multi-modal: Interviews, participant observations and field notes	“Triangulation” of data, method unclear	Themes of “Campers” 1. Kids Like Me 2. Not the Only One 3. Perspective on IBD Themes of “Noncampers” 1. Disease-centered 2. IBD and Notions of Self 3. Kids Like Me
Velsor-Friedrich et al. (2004) United States	Asthma	To explore the experiences and behaviours related to self-management of asthma in adolescents	<i>n</i> = 24 young people with asthma Age: 14-18 years Gender: 13 female, 11 male Ethnicity: Caucasian, African-American and Hispanic (numbers not provided)	Focus groups, using an interview guide	Ethnographic approach based on Morgan (1988) and Krueger (1998)	Four themes and one additional section with further sub-sections: 1. Wanting to Be Normal 2. Unpredictability of the Disease 3. Credibility of the Teen With Asthma 4. Self-Management Issues 5. Teens’ Recommendation of What Teens with Asthma Need to Know Sub-sections: <i>What Teens Need to Know; Inhaler-Related Information; Communication with Peers and Adults; Learning Styles Teens Suggested</i>

Walker et al. (2014)	Asthma	To explore the impact of school asthma management on physical activity	<p>n = 23 young people with asthma</p> <p>Age: 8-10 years</p> <p>Gender: 12 girls, 11 boys</p> <p>Ethnicity: 6 'African-American', 2 'White', 1 'More than one race', 14 'Unspecified'</p>	Semi-structured interviews, using an interview guide	Thematic and content analysis	<p>Five themes:</p> <ol style="list-style-type: none"> 1. Asthma symptoms during in-school physical activity 2. Methods to control asthma episodes during school physical activity 3. Methods to prevent asthma episodes during school 4. Limited accessibility of asthma medications 5. Negative feelings about asthma and medication use
United States						
Wang et al. (2010)	Diabetes Mellitus (Type I)	To better understand the experiences of adolescents with Type I diabetes at school	<p>n = 2 young people with Type I Diabetes</p> <p>Age: 12-16 years</p> <p>Gender: 1 female, 1 male</p> <p>Ethnicity: Not stated</p>	Semi-structured interview, using an interview guide	Hermeneutic circle, based on the hermeneutic phenomenology method.	<p>Four themes:</p> <ol style="list-style-type: none"> 1. Learning to be master of their disease 2. Learning to find ways to feel comfortable 3. Learning not to be different 4. Learning not to let others worry about them
Taiwan						

Winger et al. (2014)	Chronic fatigue syndrome	To explore the experiences of being an adolescent with chronic fatigue syndrome	n = 18 adolescents with chronic fatigue syndrome Age: 12-18 years Gender: 12 female, 6 male Ethnicity: Not stated	Semi- structured interview, using an interview guide	Phenomenological hermeneutical method	One core theme, with three subthemes: 1. “Sometimes it feels as if the world goes on without me” Subthemes: <i>On the side of life – locked in and shut out; The body, the illness and me; Handling life while hoping for a better future</i>
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inflammatory bowel disease (IBD; Salazar et al., 2014) and one on chronic fatigue syndrome (Winger et al., 2014). Children ranged from six to 18 years in age, and all studies included both female ($n = 202$) and male participants ($n = 166$). Only six studies provided information about the ethnicity of participants (Christian et al., 1997; Elliott et al., 2005; Freeborn et al., 2013; Protudjer et al., 2009; Velsor-Friedrich et al., 2004; Walker et al., 2014), and these included “White American”, “Native American”, “Caucasian”, “Asian-Canadian”, “African-Canadian”, “White Canadian”, “African American”, “Hispanic” and “More than one race”.

Although one study investigated disclosure as a facet of stigma (Benson et al., 2015a) and one study aimed to understand normalising strategies (Protudjer et al., 2009), no other studies were investigating stigma as their primary focus. Similarly, only one study had an explicit objective to understand illness perceptions (Salazar et al., 2014). Six studies aimed to explore experiences of children living with their condition (Chen et al., 2010; Christian et al., 1997; McMurray et al., 2001; Nahal et al., 2019; Wang et al., 2010; Winger et al., 2014), three aimed to explore quality of life (Elliott et al., 2005; McEwan et al., 2004; Moffat et al., 2009), and two focused on understanding factors influencing physical activity (Moola et al., 2011; Walker et al., 2014). The remaining studies had the following objectives related to understanding experiences of living as a child with a chronic health condition: understanding the challenges (Freeborn et al., 2013), the costs or rewards (Herrman, 2006), and experiences of self-management of condition (Velsor-Friedrich et al., 2004).

4.2. Quality appraisals of the included studies

A total of nineteen studies were reviewed using the CASP (1998) checklist, and two studies were excluded as a result of poor quality (Crespo-Ramos et al., 2018; Houston et al., 2000). Poor quality was determined by studies meeting less than half the checklist criteria. Ten papers were rated by both the author and another member of the research team, and the degree of agreement was assessed using Cohen’s Kappa.

There was a substantial level of agreement between the appraisals of the two raters of the papers (Landis et al., 1977), $k = .771$ $p < 0.001$. A summary of the information extracted using the CASP checklist for the seventeen included studies is provided in Table 3, with further details presented below.

4.2.1. Aims and methodology

All studies provided a clear aim, referred to the qualitative methodology being used, and provided a rationale for the research design used. All studies used purposive sampling, but only seven explicitly discussed recruitment issues such as drop-out or exclusion of data (Elliott et al., 2005; McMurray et al., 2001; Moffat et al., 2009; Protudjer et al., 2009; Walker et al., 2014; Winger et al., 2014).

All studies completed their data collection face-to-face, with 11 studies using semi-structured interviews (Benson et al., 2015a; Chen et al., 2010; Christian et al., 1997; Elliott et al., 2005; Herrman, 2006; McMurray et al., 2001; Nahal et al., 2019; Protudjer et al., 2009; Walker et al., 2014; Wang et al., 2010; Winger et al., 2014), three studies using focus groups (Freeborn et al., 2013; McEwan et al., 2004; Velsor-Friedrich et al., 2004), one study using both focus groups and semi-structured interview (Moffat et al., 2009) and two studies using a multi-modal approach which includes a semi-structured interview (Moola et al., 2011; Salazar et al., 2014). Only one study did not state the length of the interviews or focus groups (Salazar et al., 2014), with the length of time ranging from 15 minutes to two hours in the other sixteen studies. A range of locations were used: two in the participants' home (Chen et al., 2010; Nahal et al., 2019), two in their schools (Velsor-Friedrich et al., 2004; Walker et al., 2014), five in their clinics (Christian et al., 1997; McEwan et al., 2004; McMurray et al., 2001; Moffat et al., 2009; Moola et al., 2011), one in a research centre (Winger et al., 2014), one in a private room within a childrens' camp (Herrman, 2006), one in a restaurant (Wang et al., 2010) and four studies provided participants with a choice, usually between the

Table 3

Quality appraisal rating using the CASP checklist

Study	Aims	Method	Design	Recruitment	Data collection	Relationships	Ethical issues	Analysis	Findings	Value of research
Benson et al. (2015a)	✓	✓	✓	✓	✓	?	✓	✓	✓	✓
Chen et al. (2010)	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
Christian et al. (1997)	✓	✓	?	✓	✓	?	?	?	✓	✓
Elliott et al. (2005)	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
Freeborn et al. (2013)	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
Herrman (2006)	✓	✓	✓	✓	✓	?	✓	?	?	✓
McEwan et al. (2004)	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
McMurray et al. (2001)	✓	✓	✓	✓	✓	?	✓	✓	✓	✗
Moffat et al. (2009)	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
Moola et al. (2011)	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
Nahal et al. (2019)	✓	✓	✓	✓	✓	?	✓	✓	✓	✓
Protudjer et al. (2009)	✓	✓	✓	✓	✓	?	✓	✓	✓	✓
Salazar et al. (2014)	✓	✓	✓	✓	✓	?	✓	?	?	✓
Velsor-Friedrich et al. (2004)	✓	✓	✓	✓	✓	?	✓	?	?	✓
Walker et al. (2014)	✓	✓	✓	✓	✓	?	✓	✓	✓	✓
Wang et al. (2010)	✓	✓	✓	✓	✗	✓	?	✓	✓	✗
Winger et al. (2014)	✓	✓	✓	?	✓	✓	✓	✓	✓	✓

Note. Ratings: ✓ (Yes), ✗ (No), ? (Can't tell)

clinic and their homes (Benson et al., 2015a; Elliott et al., 2005; Protudjer et al., 2009; Salazar et al., 2014).

4.2.2. Relationships and reflexivity

Only seven studies stated their epistemological position (Chen et al., 2010; Herrman, 2006; Moola et al., 2011; Nahal et al., 2019; Salazar et al., 2014; Wang et al., 2010; Winger et al., 2014), whilst the remaining ten did not. Eight studies made reference to the influence of the researcher (Chen et al., 2010; Elliott et al., 2005; Freeborn et al., 2013; McEwan et al., 2004; Moffat et al., 2009; Moola et al., 2011; Wang et al., 2010; Winger et al., 2014), whereas nine did not.

4.2.3. Analysis

All studies stated the use of qualitative analysis, but there was variation in both the methodology used and the transparency of how this was reported. With the exception of one study which provided no details about the recording of data (Salazar et al., 2014), all studies transcribed the data verbatim. Five studies used thematic analysis (Benson et al., 2015a; McEwan et al., 2004; Protudjer et al., 2009; Walker et al., 2014), four used phenomenological hermeneutic methodologies (Chen et al., 2010; Nahal et al., 2019; Wang et al., 2010; Winger et al., 2014), and four used grounded theory (Christian et al., 1997; Elliott et al., 2005; Moffat et al., 2009; Moola et al., 2011). Other studies used methodology informed by the social exchange theory (Herrman, 2006), a framework approach (McMurray et al., 2001) and an ethnographic approach (Velsor-Friedrich et al., 2004). The use by Freeborn et al. (2013) of a “standard qualitative method” was less clear, although some description of the analysis process was provided. The description of Salazar et al. (2014) provided the least transparency and clarity, referring only to the “triangulation of data”.

4.2.4. Findings and value of the research

All studies provided quotes to support their themes. Most studies considered possible limitations of their research, but four did not (Herrman, 2006; McMurray et al., 2001; Moola et al., 2011; Winger et al., 2014). The final item of the CASP refers to how 'valuable' the research is, and considers factors such as whether researchers discussed the contribution of the study related to the existing literature, whether new areas of research were identified, and whether researchers discussed whether or how findings could be transferred to other populations. With the exception of one study (Protudjer et al., 2009), all studies discussed the clinical implications of their findings. Most studies made suggestions for future research, but five did not (McMurray et al., 2001; Nahal et al., 2019; Protudjer et al., 2009; Walker et al., 2014; Winger et al., 2014). Based on this, fifteen of the studies were considered to provide a 'valuable' addition to the literature, whilst two were not (McMurray et al., 2001; Wang et al., 2010).

Since the included studies were generally of a high quality, the relative quality of each paper was not considered in the process of analysis.

4.3. Thematic Synthesis

Two analytic themes of 'components of illness perceptions' and 'components of stigma experiences' and eight sub-themes were generated by thematic synthesis of the included studies. The sub-themes were based on initial descriptive themes identified in the second stage of thematic synthesis, following the first stage of line-by-line coding. The initial descriptive themes were revised and used to generate the analytic themes in the third stage of thematic synthesis. Figure 2 illustrates the final analytic and sub-themes derived from this process, and the link between these components.

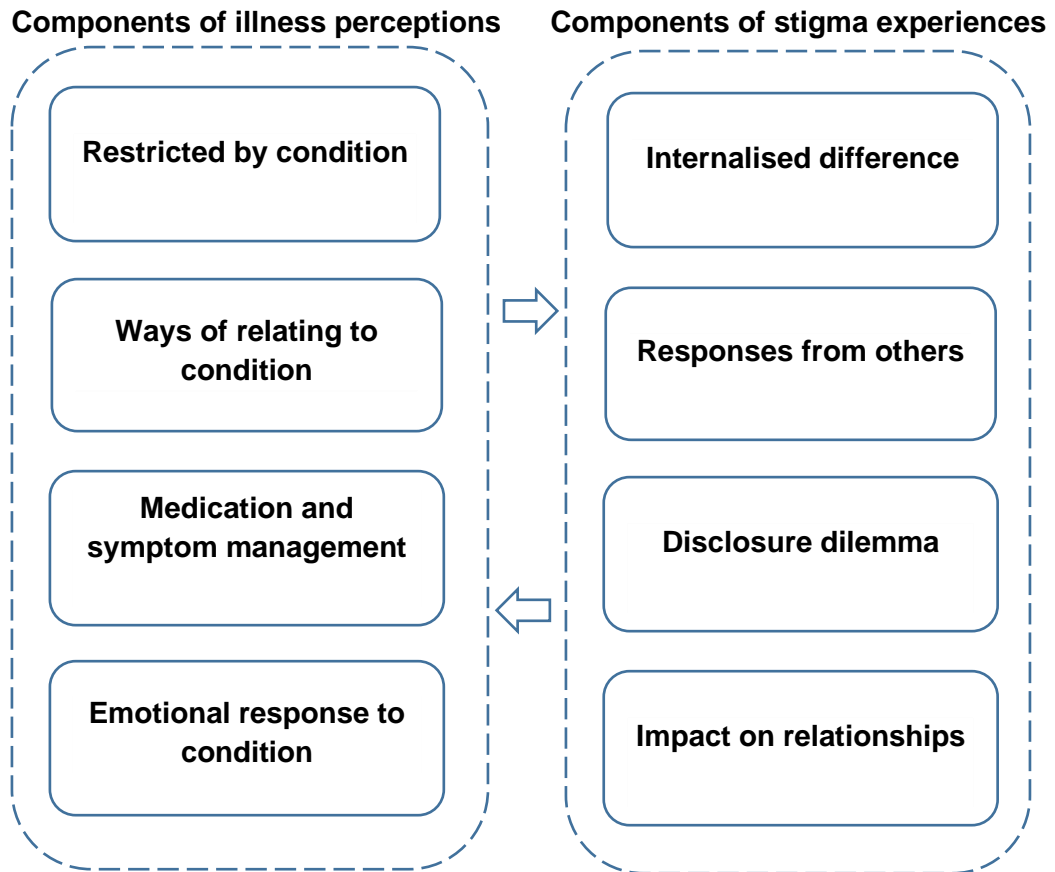


Figure 2

Analytic and sub-themes from thematic synthesis

4.3.1. Components of Stigma Experiences

Participants across all studies reflected on stigmatising experiences. These included: an internalised sense of difference; responses from others to their condition; the disclosure dilemma; and the impact their condition had on relationships.

4.3.1.1. Internalised difference

Many participants spoke about feeling different as a result of their condition. Descriptions of this included: “feeling separate”, like “I don’t belong”, “not normal”, “separated from people”, “apart from my friends”, “invisible” and “weird”. As well as not feeling normal because of their condition, young people spoke about wanting to be normal and striving for normality. For some this involved wishing for an idealised self without a health condition, whereas for others striving for normality involved trying

to engage in activities as their peers would or hiding their difference. There was also acknowledgement of the effort required in trying to be normal.

“I hate the wheelchair... I hope to get rid of it... It annoys me. I cannot be like others... I often think ‘Why me? Why am I not a normal child like others?’”
(Nahal et al., 2019)

“cause you feel like really, really, odd and you feel really, really weird that you have to sit out just because you were born with it [congenital heart disease]... you want to live a normal life, but you can’t live a normal life because you have this.” (Moola et al., 2011)

For some young people, they still perceived themselves as different, but without seeing this is as negative. As one participant described it: “I feel different, but not in a bad way” (Moffat et al., 2009). In two studies, there were also some accounts of younger children who did not feel different to their peers, by either not seeing events in their life as illness-related or instead focussing on what similarities they had with peers.

“My brothers are similar to me. The only difference is that they can walk. But I can crawl and control the wheelchair.” (Nahal et al., 2019)

4.3.1.2. Responses from others

Participants spoke about how they were treated differently by other people. Descriptions of how they believed others perceived them included “different”, “weird” and “contagious”, where the visibility of their condition or medication use was seen as a factor in marking them out as different. Peer responses to children with chronic health conditions included avoiding, excluding or rejecting them, as well as being scared of the child’s condition. Accounts of being bullied or teased were also presented in most studies.

“At school, the boys in my year and other boys in other years pick on me, they call me names and things just because I’ve got epilepsy, I’ve had that since I started school” (McEwan et al., 2004)

There was also evidence of others using stereotypes to inform how they responded to participants. Young people described others seeing them as contagious, with consequences of social exclusion or rejection. Others were seen as being frail, weak or fragile, which also provided a barrier to involvement in physical or social activities. A lack of understanding by others of their condition was frequently cited by participants as informing how peers and teachers responded to them. In some instances, this led to participants having to educate others about their conditions.

“I don’t tell my good friends the truth, they would be scared. I have no friends, they are afraid of being infected by me. Although I explained to them, they were still doubtful...” (Chen et al., 2010)

“...like some people don't really like to sit beside me... In case I... in case I would get a seizure or something like that.” (Benson et al., 2015a)

Alternative accounts to these negative responses, stereotypes and lack of understanding were described in the context of being around children with the same condition. This helped participants to feel understood, to feel a sense of belonging, to feel less alone and to learn from each other. The only exception to this was in a participant group in one study who had chosen not to attend IBD camp, although the majority of these children also recognised the benefits of being around children with the same condition.

“It almost makes me feel normal. I said, ‘Yes! This is great; I can teach you everything I know!’ (Freeborn et al., 2013)

“It’s just kind of depressing [going to IBD camp]. They’re all going to be sitting up there talking about how “we’re sick” (Salazar et al., 2014)

4.3.1.3. Disclosure dilemma

The dilemma of whether to disclose their condition came up in most studies, with children dealing with the dilemma in different ways. Concealment of their condition or hiding the use of medication was commonly cited. The perceived risks of disclosing their condition included: stigma, being treated and perceived differently, being bullied, not being understood and being embarrassed. Some of these fears appeared to be particularly present in the context of romantic relationships in adolescents. Benefits of disclosure included feeling safer, more supported in the management of their condition, and support from friends and family. Other factors influencing disclosure dilemmas included parental attitudes towards concealment, whether children found it hard to talk about their condition, or their own illness perceptions.

“No [in response to telling others about asthma]... Because, um, they might be calling you Asthma Boy or something, making you names, so... or who knows what they’ll... I would never tell them that.” (Walker et al., 2014)

Some children opted for selective disclosure, by disclosing their condition to some people but concealing it from others. Others only disclosed symptoms, rather than the label of their medical diagnosis. If children were choosing to disclose, they generally reported doing so to their close friends, although some did not tell anyone outside of their immediate family. Participants were more reluctant to disclose to certain teachers for fear of being treated differently, or to peers whom they did not trust to not tell other people or peers who they believed would bully them if they found out.

“I’ve only told one person. I just think I want a couple to know but I don’t want everyone to know in case they run around the street telling everyone I’ve got epilepsy. I wouldn’t want thousands of people to know” (McEwan et al., 2004)

However, the costs of concealment were also discussed. It was acknowledged by participants that this could impact on social relationships, affect their safety in managing their condition and also have an emotional impact on them.

“Well, everybody I've told, like they break up with me, and so I don't want to tell him because I really like him... because if I tell him, he is going to break up with me... So it doesn't hurt me, but a little it does, because I want to tell him.” (Christian et al., 1997)

4.3.1.4. Impact on relationships

In many of the studies, participants reflected on the social isolation resulting from having a condition. The reasons cited for this included both internal and external factors. Internal factors contributing to social isolation included: a lack of self-confidence, having nobody to identify with, feeling different from peers, embarrassment, feeling self-conscious, feeling forgotten, and avoiding contact with peers due to fears of negative attitudes of others. External factors given for social isolation included: being bullied, being avoided by peers due to stereotyped fears of being contagious, restrictions imposed by parents and teachers, not being able to keep up with peers, and having nobody to identify with. Social isolation led to a sense of loneliness for several participants.

“I have thought that... I don't really belong... yeah like when I say I don't belong here I feel like... I should have never been born... sometimes I've thought that I could just be invisible and nobody would really care... or I could not be here and nobody would notice and I just felt basically like a nobody” (Elliott et al., 2005)

For participants who did have close friendships, this was generally seen as protective. Close friendships were cited as helping participants adhere to their medication,

facilitate disclosure to others, provide validation and provide social support. Friends were also seen to provide safety by being able to help them with a medical emergency if required, but also to stick up for them against bullies.

I just hated getting my medicine... My friend, she's always, take your medicine, she's always like, you know, take it, no one will ever see you take it. It's not real noticeable, just reach into your purse and then put it in your mouth, take a drink real quick, no one really notices. (Christian et al., 1997)

Interviewees also spoke about the impact their condition could have on familial relationships. Several children discussed the impact that the condition had on their family members, including how their medication, illness management or emotional responses impacted on their parents and siblings. Some children felt their condition was a burden to parents and had an impact on parental relationships. Conversely, other children spoke about how their condition had helped them become closer with family members.

"My parents are divorced... it's the diabetes and the stress" (Herrman, 2006)

4.3.2. Components of Illness Perceptions

Participants across all studies reflected on how they perceived their chronic health conditions. The four component sub-themes were: feeling restricted by their condition; ways of relating to their condition; medication and symptom management; and the emotional response to their condition.

4.3.2.1. Ways of relating to their condition

In most of the studies, children spoke about the way in which they negatively related to their condition. Participants commented on how they did not want to have a condition and a sense of injustice about this. Some used the word "hate" to describe how they felt about their symptoms or condition. Participants also discussed the way

in which they viewed their bodies in relation to their condition. Accounts of how children related to their chronic health conditions included seeing their bodies as “broken” and “strange”, and not being able to count on a body which felt out of their control. A link with stigma was apparent here too, with perceptions of ‘normality’ influencing how children relate to their condition.

“Yes... or you don't feel as... an adolescent anymore. Feel [...] that... you are stuck; you are stuck in your body or something” (Winger et al., 2014)

‘I would be a clean girl... like have nothing wrong with me... fixed, not broken... normal, just a normal person’ (Elliott et al., 2005)

However, there were also accounts of more positive feelings about their conditions. There was acknowledgement by some participants of possible rewards of having a chronic health condition, such as getting more attention and receiving certain concessions at school. Others reflected on the coping strategies they used to make themselves feel better. Some children spoke about coming to accept their condition, and how things “could be worse”.

Participants spoke also about their identities. While some children did not see their condition as part of their identity, others did. For those who did, some did not want to be characterised or defined by their condition. Conversely, others reflected on a process of developing a new sense of self through the experience of having a chronic illness.

4.3.2.2. Restricted by the condition

Participants spoke about feeling restricted by their chronic health condition in a number of ways. Several participants reflected on feeling less able than their peers without chronic health conditions, discussing this in the context of physical exercise, social confidence and academic achievement. Missing school and break times were

seen as having consequences on academic ability and confidence. Feeling less able in physical exercise was also noted several times, where participants described becoming tired more quickly, having to put in more effort or performing less well than peers. Concerns about or experiences of missing out on social activities was also frequently mentioned. This included accounts of missing break times at school, school absences, not being able to out with friends or missing sleepovers.

“Why do we have to do a sport today? And I’m not any good at basketball. I can’t dribble, and I’ll feel like a loser cause I’m going to mess up all the time, I’m not going to get any baskets. Yah-just feeling left out... there’s a group of six girls that were really good at every sport. And I’d be like “oh I wish I could, you know?” At least be good at one – it would make me feel a little better.”
(Moola et al., 2011)

“I can’t do sleepovers...’cause of having to check and going low [blood sugar]... I really want to go to a sleepover.” (Herrman, 2006)

Parents were also perceived by some participants as imposing restrictions on them. This included parental monitoring of medication or illness-management, restricting unsupervised activity and parents being seen as overprotective. However, there were also concerns about how they would manage in the future without the support of their parents.

“Mum’s a bit over-protective sometimes. If I’m going out she asks me all the normal questions like where am I going – but then it’s like ‘Don’t push yourself too much’ and I already know. It gets a bit irritating after a while”
(McMurray et al., 2001)

However, some children did not perceive their condition as restrictive. Whilst most of these children acknowledged potential barriers, they spoke about either focussing on other activities or working around barriers to certain activities.

“I would say you have to know your boundaries: where you can go and where you can’t. If you have asthma, it’s going to affect you, but you can usually still do the things you can.” (Velsor-Friedrich et al., 2004)

4.3.2.3. Medication and symptom management

Children did not just speak about their condition, but also about the medication and management associated with this. Medication was described as being a burden, time-consuming, creating undesired side effects, and having a negative emotional impact. Symptom management strategies such as having to check blood glucose levels in diabetes was similarly described as inconvenient, painful and annoying. Medication was also seen as highlighting difference between participants and their peers, with accounts of children trying to hide the use of their medication as a result. There were also comments about medication adherence being affected by concerns about how others might respond, despite an awareness across some of the participants about some of the potential costs of not adhering to their medication or management plans. This indicates how stigma experiences and illness perceptions can intersect around medication.

“He [boyfriend] never asked me any questions, and I never really took my medicine in front of him... When we went out to eat I never really took them with me.” (Christian et al., 1997)

“When I am home and not feeling well, I will test my sugar without considering anything. If I want to test, I test, but at school I feel some stress. I don't like my classmates watching... Testing in the restroom is inconvenient. One time, I

accidentally dropped my meter into the toilet... Sometimes I skip the test... Now, when I test my sugar in the classroom, I look to make sure no one is watching me, and then I will do a quick test on my thighs. I would never put the meter on my desk.” (Wang et al., 2010)

4.3.2.4. Emotional response to their condition

Generally, children’s emotional narratives about their experiences of having a chronic health condition in these studies were centred around distress, although one paper referred to participants reports of being ‘happy’. A wide range of emotions were expressed: experiences around fear, anxiety or worry were most frequently cited by interviewees. For some, worries were related to the condition itself: the fear of having symptoms, doing something to exacerbate symptoms, future medical interventions, and for some, a fear of dying. For others, worries were related to stigmatisation from other people: being bullied, people finding out about their condition or being rejected. These worries were also frequently discussed in the context of feeling embarrassed. Adolescents discussed worries about their future as adults, including being discriminated against or not having people to support them with their condition once they left home.

“Managing university studies alongside a serious health condition and restrictions in mobility can be a daily struggle. I will not be able to study at university, or to work and get married. I’m afraid of what will happen to me if my mother is no longer able to care for me ... who will help me in this miserable life?” (Nahal et al., 2019)

Anger, frustration and annoyance were also commonly referred to by participants. Children described feeling annoyed about being restricted in their activities, having to take medication and having to experience symptoms. The negative reactions of

others were also a source of anger and frustration, particularly in response to being treated differently or not being understood.

“[E]verything [makes me angry]... when I’m in a... angry mood, I can get along really bad, with mum and dad, especially dad... they [parents] say all kinds of things like... no, I can’t leave you swimming here because, um, the other parents don’t know your medical history... it makes me feel... angry, frustrated”

(Elliott et al., 2005)

Participants also spoke about their condition as making them sad, depressed or upset. Children felt sad about having the condition, but also as a result of social comparisons of feeling different and less able than others. There were also accounts of how this led to suicidal thoughts in some participants.

4.3.3. Link between ‘Components of Stigma Experiences’ and ‘Components of Illness Perceptions

Although the sub-themes have been grouped within distinct constructs of stigma and illness perceptions, Figure 2 shows bidirectional arrows to represent how these may interact with each other. For example, within illness perceptions, the sub-theme of ‘medication and symptom management’ related to stigma experiences. The use of medication was seen to visibly highlight an area of difference, and stigma was cited as a reason for non-adherence to treatment plans. Peer support in relationships where stigma was felt to be absent was cited as increasing medication adherence. Similarly, sub-themes within stigma experiences led to ‘emotional responses’, a sub-theme of illness perceptions, suggesting that the stigma could also influence illness perceptions. For example, accounts of bullying and teasing within the ‘responses from others’ sub-theme and social isolation within the ‘impact on relationship sub-theme’ were described alongside emotional responses of sadness, worry and embarrassment about their condition. ‘Emotional responses to their condition’ as a

sub-theme was also described in the context of the disclosure dilemma. The arrows in the diagram in Figure 2 are therefore bidirectional, indicating that stigma experiences and illness perceptions both relate to and influence each other. However, given the complexity of some of these relationships, these arrows represent a tentative link of how these different sub-themes within stigma and illness perceptions may interact with each other.

“Yes and I try to keep it private...Because it sort of makes me sad...Because I don't want to have it... Because it just...it doesn't really...I don't really like it...No because I would feel embarrassed, I would feel upset, I would feel all the bad things.” (Benson et al., 2015a)

“Um, I feel, I feel sad [about using pump in front of classmates]. Because I'm scared that everybody's going to tease me.” (Walker et al., 2014)

5. Discussion

5.1. Summary of Findings

The aim of this review was to better understand perceptions of illness and physical health stigma experiences in children with chronic health conditions, and the inter-relationship between the two. A thematic synthesis based on the stepwise approach by Thomas et al. (2008) was used to generate two analytic themes and eight sub-themes from the data of 17 qualitative studies.

5.1.1. Stigma experiences

Within the analytic theme of 'components of stigma experiences', four sub-themes were identified: an internalised sense of difference; responses from others to their condition; the disclosure dilemma; and the impact their condition had on relationships. These sub-themes correspond to the existing stigma literature. In the Child Stigma Scale developed by Austin et al. (2004) to measure stigma in children with epilepsy,

questions are similarly based around areas such as feeling different, concerns about the responses of others and whether they attempt to keep their epilepsy a secret. However, while the sub-themes are linked, they relate to different aspects of a stigmatising experience.

The differentiation of 'internalised difference' and 'responses from others' in this review as distinct components of stigma are aligned with ideas in the wider stigma literature. *Public stigma* is a concept used to describe the responses of others (Bos et al., 2013), while the internalisation of these stigmatising experiences is described by the term *self-stigma* (Corrigan et al., 2002).

Similarly, the 'disclosure dilemma' of whether to disclose or conceal in stigmatised identities is increasingly being recognised as a key component of stigma generally (Quinn et al., 2017), and also within children with chronic health conditions (Benson et al., 2015b; Kaushansky et al., 2017). In this review, children more frequently spoke about choosing to hide their chronic health conditions in the context of the 'disclosure dilemma'. A review of concealment across different stigmatised identities highlighted social isolation, preoccupation with concealment and negative affective responses as possible psychological implications of concealment (Pachankis, 2007). The 'impact on relationships' sub-theme recognises another potential consequence of internal and external stigma experiences, and is particularly relevant in the context of the negative consequences of social rejection (Pittman et al., 2007; Sandstrom et al., 2004; Strauss et al., 2007).

Making distinctions about different components of stigma rather than understanding it as one entity is important, because it has clinical implications for how children with chronic health conditions can best be supported. If children with chronic health conditions are found to consistently experience external stigma through the negative responses from others, then future research should build on existing studies

investigating societal perceptions of children with chronic health conditions (e.g. Ani et al., 2011; Banko, 1999; Hayes et al., 2013; Masnari et al., 2013b) and consider relevant systemic interventions. Conversely, if children with chronic health conditions are experiencing stigma more as an internalised sense of difference through self-stigma, then interventions supporting the stigmatised individual with this may be more appropriate (Heijnders et al., 2006). For example, Corrigan and colleagues have developed a stigma intervention based on disclosure decisions for individuals with mental health problems (Corrigan et al., 2013) with the aim of reducing self-stigma. Therefore, in order to better understand stigma experiences in children with chronic health conditions, further research is required to investigate how different components of stigma are experienced by children with chronic health conditions and how these components relate to each other. This could have clinical implications for how children with chronic health conditions can best be supported with stigma experiences.

5.1.2. Illness perceptions

Within the analytic theme of 'components of illness perceptions', four sub-themes were identified: ways of relating to their condition; feeling restricted by their condition; medication and symptom management; and the emotional response to their condition. Many participants spoke about the negative ways in which they related to their condition, including wishing they did not have the condition, a sense of injustice, and feeling let down by their bodies. Being 'restricted by the condition' was also identified as a sub-theme, with children experiencing their condition as a barrier to socialising, physical activity and other desired activities. While these components are similar to those included in measures of illness perceptions such as the CATIS (Austin et al., 1993), 'medication and symptom management' is less apparent in the literature. However, in this review it clearly emerged as an area in which children were having negative experiences, both related to the adherence to medication and also as a factor affecting their visibility as having a stigmatised identity. Future research should

consider the impact of medical interventions and lifestyle changes required for symptom management in children's illness perceptions and experiences, which is currently absent in the cited quantitative measures (Austin et al., 1993; Austin et al., 2004).

The final sub-theme of the 'emotional responses to their condition' included accounts of experiencing fear, anger and sadness. This aligns with previous research which has shown a link between negative illness attitudes and depressive symptoms, stress, anxiety (Austin et al., 2006; Austin et al., 1993; le Coq et al., 2000; LeBovidge et al., 2005; Ramsey et al., 2013). However, researcher bias may have been present in the qualitative studies included, where an assumption of distress from researchers could have elicited responses of negative emotions. Given that only eight of the 17 studies clearly reflected on their relationship to the participants and none of these cited the assumption of distress as a potential area of researcher bias, this should be taken into account when interpreting findings related to participants' emotional responses.

In the adult physical health literature, the common sense model of illness representation suggests that individuals hold both cognitive and emotional perceptions of their health condition (Diefenbach et al., 1996; Hagger et al., 2003). The illness perception sub-themes in this review could be similarly conceptualised, with 'emotional responses' as an emotional illness perception and the other three sub-themes as cognitive illness perceptions. Given the potential malleability of cognitive illness perceptions, a better understanding of these could be used to inform psychological interventions offered to children who are experiencing distressing emotions as a result of their chronic health condition. For example, cognitive-behavioural therapy interventions could be used to support children who feel restricted by their condition through behavioural activation or those who have negative ways of relating to their condition by reappraising relevant cognitions (e.g. Christie et al., 2005).

5.1.3. Stigma experiences and illness perceptions

Although stigma experiences and illness perceptions represent two distinct constructs in the existing literature, the findings in this review align with evidence that they are related (Austin et al., 2004; Funderburk et al., 2007). For example, as part of children's illness perceptions, the requirement to take medication or manage their condition was seen as burdensome by many participants. Although this was partly due to issues like the side-effects of medication, the act of taking medication as highlighting difference was related to stigma experiences. Similarly, there appeared to be links between component sub-themes of both constructs. The 'impact on relationships' was in part related to being 'restricted by the condition': component sub-themes of stigma experiences and illness perceptions respectively. This has clinical implications because if stigma and illness perceptions are related, interventions focussed on one construct could also affect the other. Although the relationship between self-stigma and illness perceptions has been established in children with epilepsy (Austin et al., 2004; Funderburk et al., 2007), future quantitative research could be used to confirm how the findings in this review apply in other chronic health conditions. Further research around relationships between the different components of stigma and illness perceptions would also help provide a better understanding of children's illness experiences and guide the focus of clinical interventions.

5.2. Quality of the included studies

The CASP checklist was used to evaluate the quality of nineteen eligible studies. Seventeen studies were deemed of satisfactory quality to include, and two were excluded based on the quality analysis. The included studies varied in quality and in the transparency of their methodological procedures, although they were considered to be of a high enough standard that relative quality was not used to prioritise different studies within the analysis. The CASP item of 'consideration of the relationship between researchers and participants' was most often absent in the descriptions of

the included studies. The quality of studies could have been improved by researchers reflecting upon their assumptions, possible bias, and by being more explicit in their epistemological position. Similarly, the 'rigorousness of data analysis' was another CASP item where the descriptions of some studies were unclear, whereby the quality of studies could have been improved by describing their analysis with more clarity. These limitations related to the quality of the studies should be taken into account when interpreting the findings in this review.

5.3. Limitations

First, although it was felt that conceptual saturation was reached in the thematic synthesis, the method used to identify studies was based on an exhaustive sampling strategy more commonly used in meta-analyses. There a debate regarding the best sampling approach in qualitative synthesis studies within the wider literature (Toye et al., 2014). In this study, the rationale for exhaustive sampling was based on being inclusive in a developing area of the literature, but there was a risk of not reaching conceptual saturation. Second, although multiple databases were searched, relevant papers may have been missed. Due to the nature of qualitative research prioritising the voice of participants, the word 'stigma' was not often used in titles and abstracts of studies. Children may have been more likely to use words or phrases such as "different" or "not normal" to describe stigma experiences. Therefore, by only using variations of the word "stigma" as part of the search strategy, relevant studies may have been missed. Attempts were made to overcome this by using ancestry searching of bibliographies of potentially eligible studies and of relevant review papers. Third, excluding non-English studies and grey literature means there is an increased risk of publication bias and also risks exclusion of the experiences of non-English speaking cultures. Although one of the studies was based in Palestine (Nahal et al., 2019), it was difficult to make inferences about cultural factors when all other studies were based on Western and English-speaking samples. Fourth, most studies did not have

the explicit aim of investigating stigma and illness perceptions, with findings related to these constructs being incidental. This could have limited the exploration of these constructs and led to a bias towards what was being investigated (for example, disclosure in the (Benson et al., 2015a) study), and means some caution is required in the interpretation of these findings.,

5.4. Future Research

One of the findings of this study was that medication and symptom management emerged as a sub-theme within children's illness perceptions, despite this currently being absent in the cited quantitative measures (Austin et al., 1993; Austin et al., 2004). Therefore, future research could involve the development of measures which more broadly represent children's illness perceptions, or further qualitative research about how medication and symptom management affects children across a broader range of health conditions. Secondly, the perspectives of parents, professionals and peers were excluded in this study, in order to prioritise the perspective of the children themselves. However, gaining a better understanding of how stigma is perceived by people around the child is also important, and future research focussed on stigma within children's systems would be useful. Thirdly, the findings of this study suggest that there may a relationship between stigma and illness perceptions and within the components identified within these constructs, but the bidirectional relationship between these is tentatively proposed in this study due to the complexity of this. Future research could explicitly investigate this, by asking children with a range of chronic health conditions about their experiences of stigma and illness perceptions.

5.5. Conclusions

By examining stigma experience and illness perceptions as described in the existing qualitative literature, this review has provided insight into how children see their chronic health conditions and how they believe others perceive them. Despite this being an emerging area of the literature, this review has identified different component

parts of stigma and illness perceptions, and how these might be related to each other. This has clinical implications for how children with chronic health conditions can best be supported by identifying the possible targets of psychosocial interventions, and provides the foundations for future research.

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Part 2: Empirical Paper

Stigma, Concealment, Illness Perceptions and Psychosocial Difficulties in Children with Physical Health Conditions and Their Parents

1. Abstract

Aims: The overall purpose of this study was to understand the relationships between physical health stigma, concealment, illness perceptions, and psychosocial difficulties in children with physical health conditions and their parents. Further objectives were to identify which of these factors predicted children's psychosocial difficulties, and which factors predicted concealment.

Method: A cross-sectional survey was completed by 61 child-parent pairs attending dermatology or urology outpatient clinics in a London paediatric hospital. Children and parents completed validated measures of stigma, concealment, illness perceptions, and children's psychosocial difficulties.

Results: Correlational analyses revealed that in children, stigma was associated with concealment and illness perceptions, both with a large effect size. Children's psychosocial difficulties were associated with child stigma and parent stigma, both with a medium effect size, and also with illness perceptions, with a small effect size. Multiple hierarchical regression analyses found that both child and parent stigma independently predicted children's psychosocial difficulties (a composite of emotional and peer problems). Child stigma predicted child concealment, and parent stigma predicted parent concealment. However, there was no relationship between any child and parent-rated factors.

Conclusions: The stigma perceptions of both children with physical health conditions and their parents need to be taken into account in the context of understanding the child's psychosocial difficulties, and families should be supported in making decisions about concealment. As child and parent factors were not associated with each other, wider influences on children's stigma experiences should also be considered.

2. Introduction

The estimated prevalence of chronic health conditions in children is between 13-27% and is increasing (Van Cleave et al., 2010). Evidence of a relationship between physical and mental health in this population is well-documented: children with physical health conditions are at a greater risk of mental health problems and psychological distress (Ferro et al., 2015; Hysing et al., 2007), and children with mental health problems are four times more likely to be in poor physical health than those without (Green et al., 2005). However, the wide range of illness-related factors affecting psychosocial difficulties in children with health conditions are not clearly understood. A prioritisation of children's physical health has been found to lead to an inadvertent neglect of their mental health needs (Bennett et al., 2015). Therefore, an understanding of factors contributing to psychosocial difficulties in children with physical health conditions could aid the provision of more well-rounded care.

2.1. Stigma

Stigma is associated with an area of 'difference', where deviation from societal norms is seen to discredit individuals (Goffman, 1963). Stigma is apparent in direct acts of discrimination such as hostility, but indirect discrimination also occurs through status loss and social exclusion (Link et al., 2001). The way in which stigma is experienced by individuals can be categorised into different forms. *Public stigma* refers to how the general population views and responds to stigmatised individuals, whereas *perceived stigma* refers to how stigmatised individuals understand others to see them (Bos et al., 2013). The internalisation of these attitudes by the stigmatised individuals is described as *self-stigma* (Corrigan et al., 2002), whereby they come to believe these stigmatised beliefs about themselves. Stigma has been found to have a number of negative psychological consequences, including reduced self-esteem, increased psychological distress, and reduced treatment seeking (Corrigan et al., 2006; Jennings et al., 2015; Quinn et al., 2009).

In the adult stigma literature, Quinn et al. (2014) proposed that the level of 'outness' and how individuals related to their stigmatised identity (through 'salience' and 'centrality') were also important in understanding stigma experiences and psychological distress. These ideas are considered here through the concepts of 'concealment' and 'illness perceptions', within the context of stigma and psychosocial difficulties in children with physical health conditions and their parents.

2.2. Stigma in children with physical health conditions

Children with physical health conditions are subject to physical health stigma (hereafter referred to as stigma), according to reports from children and families. Estimated stigma prevalence is high, with over half of adolescents with diabetes reporting stigma in one study (Brazeau et al., 2018). Social stigmatisation was rated as one of the worst parts of having epilepsy (Vanstraten et al., 2012), and stigma has been found to account for increased psychological distress, increased depressive symptoms, reduced self-esteem, and a lower treatment adherence (Austin et al., 2004; Shah et al., 2015; Taft et al., 2009). In qualitative studies, children have described stigma experiences as including social isolation, bullying, being seen as contagious, and being treated differently (Chen et al., 2010; Elliott et al., 2005; McEwan et al., 2004). The negative social consequences of stigma may be particularly salient in middle childhood and adolescence, as peer acceptance comes to be an increasing priority (Pittman et al., 2007) and when identity development is occurring through biopsychosocial changes (Eccles, 1999).

Parents of children with health conditions also report experiences of stigma. *Parental perceived stigma* refers to how much parents believe their children are stigmatised, whereas *courtesy stigma* involves parents being stigmatised through association with their child (Birenbaum, 1970). Understanding parental stigma experiences could be important for a number of reasons. First, parents often act as the 'gateway' to services (Chavira et al., 2017), making choices about which support their children need to

access. Parental stigma has been associated with reduced help-seeking in mental health and neurodevelopmental settings (DosReis et al., 2010; Turner et al., 2010), and could therefore influence the type of input children receive from services. Second, as with stigma experiences reported by children, there is an association between higher levels of parental perceived stigma and higher levels of their children's depressive symptoms epilepsy (Carlton-Ford et al., 1997). This indicates that parental stigma could also provide insight into emotional difficulties in their children. Finally, in children with epilepsy there is emerging evidence of what has been referred to as '*stigma coaching*' (Jacoby & Austin, 2007; Benson et al. 2016). Here, the stigmatised identity is modelled by parents as not being spoken about or shameful, leading to a conscious or unconscious transmission of stigma-related attitudes or behaviour. By asking both children and their parents about their stigma experiences in relevant self-report measures, this study sought to investigate whether stigma transmission occurred between parents and children in physical health conditions beyond epilepsy, and whether stigma was associated with the level of children's psychosocial difficulties.

2.3. Concealment of physical health conditions

Individuals with a stigmatised identity face a choice of whether to disclose or conceal their stigmatised attributes to others. This 'disclosure dilemma' is increasingly being recognised as a part of stigma experiences, whereby the concealment of a stigmatised attribute is used to prevent the negative consequences associated with stigma (Quinn et al., 2017). This dilemma has been described as important by children with epilepsy and their parents (Benson et al., 2015b), and the reasons for choosing whether to disclose or conceal are complex. Concealment has been described by children as being protective, with perceived concerns about disclosure including fears of being bullied, loss of relationships, and being seen as different (Christian et al., 1997; Freeborn et al., 2013; Moola et al., 2011). Several studies have described

children choosing a strategy of selective disclosure, such as by disclosing their condition to close family members but concealing it from certain peers (Benson et al., 2015a; Kaushansky et al., 2017; Wang et al., 2010).

Although children may choose to conceal their health condition as a protective strategy, there is evidence of negative consequences of concealment. In a review across different stigmatised identities, possible psychological implications of concealment included social isolation, preoccupation with concealment and negative affect (Pachankis, 2007). Children with health conditions have also reported adverse consequences associated with concealment in the qualitative literature, including negative impacts on social relationships, lack of support from others in management of conditions, and also the negative emotional impact of having to hide their condition (Christian et al., 1997; Moola et al., 2011). Social support can be protective against depressive affect (Luo et al., 2017), and negative social consequences may be particularly pertinent as children begin to prioritise peer acceptance during their transition into adolescence. However, a review of disclosure decisions in epilepsy concluded that there was insufficient evidence around the consequences on emotional difficulties (Benson et al., 2015b), despite this population being the most well-researched on the topic of concealment. This study therefore aims to investigate how concealment relates to psychosocial difficulties and stigma.

Parents are also involved in making decisions about the concealment of their child's health condition, and there is evidence to suggest that parental concealment is implicated in 'stigma coaching'. Parental concealment has been associated with higher levels of child stigma and concealment (Benson et al., 2016; Ryu et al., 2015), and with reduced disclosure outside of the nuclear family in children with epilepsy (Benson et al., 2015a). The aforementioned role of parents as a 'gateway' to services for their children also applies here (Chavira et al., 2017), where concealment of a health condition or related symptoms could pose a barrier for support. There are also

potential broader implications of concealment for stigma: if children are being encouraged to hide their health conditions, a lack of visibility and representation of these experiences in the public could maintain public stigma. Therefore, this study also aimed to identify which factors predicted concealment, particularly in relation to child and parent-rated stigma.

2.4. Illness Perceptions

Another element of how children experience their health conditions is their illness perceptions. This encompasses the attitudes, beliefs and feelings children have about their conditions, and also whether these are positive or negative (Ramsey et al., 2016). Although the concept of illness perceptions in the literature of children with physical health conditions is broadly similar to how it is understood in the adult literature within frameworks such as the common sense model (Diefenbach et al., 1996), much of the quantitative literature is based on the use of measures such as the Child Attitudes Towards Illness Questionnaire (CATIS; Austin et al., 1993). This measure includes questions around the emotions the child experiences related to their condition, whether they believe their condition prevents them from doing activities, and whether they feel good or bad about having a medical condition. Positive illness perceptions appear to be protective, and are associated with lower mental health symptoms and improved health outcomes (LeBovidge et al., 2005; Murphy, 1974). Conversely, negative illness perceptions have been linked to stress and depressive symptoms (Austin et al., 2006; le Coq et al., 2000; Wagner et al., 2008). Illness perceptions have also been linked to broader psychosocial factors: negative illness perceptions in epilepsy have been linked to poorer academic attainment, and to the use of coping strategies which have been labelled as 'maladaptive' in managing their condition (Austin et al., 1998; Austin et al., 1991). Finally, illness perceptions have also previously been linked to stigma in children (Austin et al., 2004). However, nearly all of these studies are based on children with

epilepsy, and may not replicate to the broader population of children with physical health conditions. This study aims to investigate how illness perceptions are related to psychosocial difficulties in children with health conditions, in the context of stigma experiences.

2.5. Rationale and Aims

The way in which someone relates to their stigmatised identity, and whether they decide to disclose or conceal it, have been identified in the adult stigma literature as important factors to consider alongside stigma and emotional difficulties (Quinn et al., 2014). For children with health conditions, the influence of parents as 'stigma coaches' have also been highlighted in children with epilepsy (Benson et al., 2016; Jacoby et al., 2007; Ryu et al., 2015). Therefore, the overall purpose of this study was to understand the relationship between stigma, concealment, illness perceptions, and psychosocial difficulties in children with physical health problems and their parents.

The 'disclosure dilemma' has been highlighted as an important and difficult decision for children with health conditions (Benson et al., 2015b). Although concealment of a health condition has been described by some children as protective against stigma from others, potential negative consequences include social isolation, lack of support, and emotional difficulties (Christian et al., 1997; Moola et al., 2011; Pachankis, 2007). Therefore, this study sought to identify predictors of concealment in children and parents.

Children with health conditions are at an increased risk of experiencing mental health problems (Ferro et al., 2015; Hysing et al., 2007), and stigma has been identified as a factor influencing emotional difficulties (Taft et al., 2009). Previous studies have found psychosocial difficulties to be associated with: child stigma in epilepsy (Austin et al., 2004), child illness perceptions in arthritis (LeBovidge et al., 2005), parent stigma in epilepsy (Carlton-Ford et al., 1997), and with concealment in stigmatised

adults (Quinn et al., 2017). However, to the author's knowledge there are no existing studies which consider the relative contribution of these factors on the psychosocial difficulties of children with a physical health condition.

Therefore, this study set out to address the following research questions:

1. What is the relationship between child and parent rated factors of stigma, concealment and illness perceptions?
2. Which child and parent factors predict concealment of the child's physical health condition?
 - 2a) Do child stigma, parent stigma and parent concealment predict concealment in children?
 - 2b) Does parent stigma predict parental concealment?
3. Which child and parent factors (stigma, concealment and illness perceptions) predict children's psychosocial difficulties?

3. Method

3.1. Participants

A convenience sample of participants were recruited from the dermatology or urology outpatients' services at a national paediatric hospital in London, United Kingdom to represent a small range of health conditions. Based on this, the inclusion criteria were as follows: 1) children aged 8-14 years old with a dermatological or a urology condition; 2) attending a dermatology or urology outpatient service with their main custodial parent or guardian during the data collection period; 3) the child and parent having sufficient English language proficiency to complete the questionnaires independently. Participants were excluded if: 1) the child was acutely unwell; 2) if either the child or parent in a child-parent pair did not want to participate.

A total of 242 child-parent pairs were identified and invited to take part. The flowchart in Figure 1 outlines the stages of recruitment and data collection, including the total number of child-parent pairs who took part, attrition rates, the transition to remote data collection as a result of Covid-19, and reasons given by parents for declining participation. The final sample comprised of 61 children with physical health conditions and their parent/caregiver (response rate = 25%). Data was collected from 49 participants face-to-face (80.3%) and 12 participants remotely (19.7%).

3.2. Design

This study used a non-experimental, correlational, cross-sectional design with two groups: children with physical health conditions and their parents. Parents and children were asked to complete a range of published scales, and parents also completed an additional survey about the demographic and medical background of the child. The child-rated variables were: child stigma, child concealment, and illness perceptions. The parent-rated variables were: parent stigma, parent concealment, and children's psychosocial difficulties. The relationship between these child and parental variables were considered, with a focus on how they might relate to: 1) children's psychosocial difficulties, and 2) concealment.

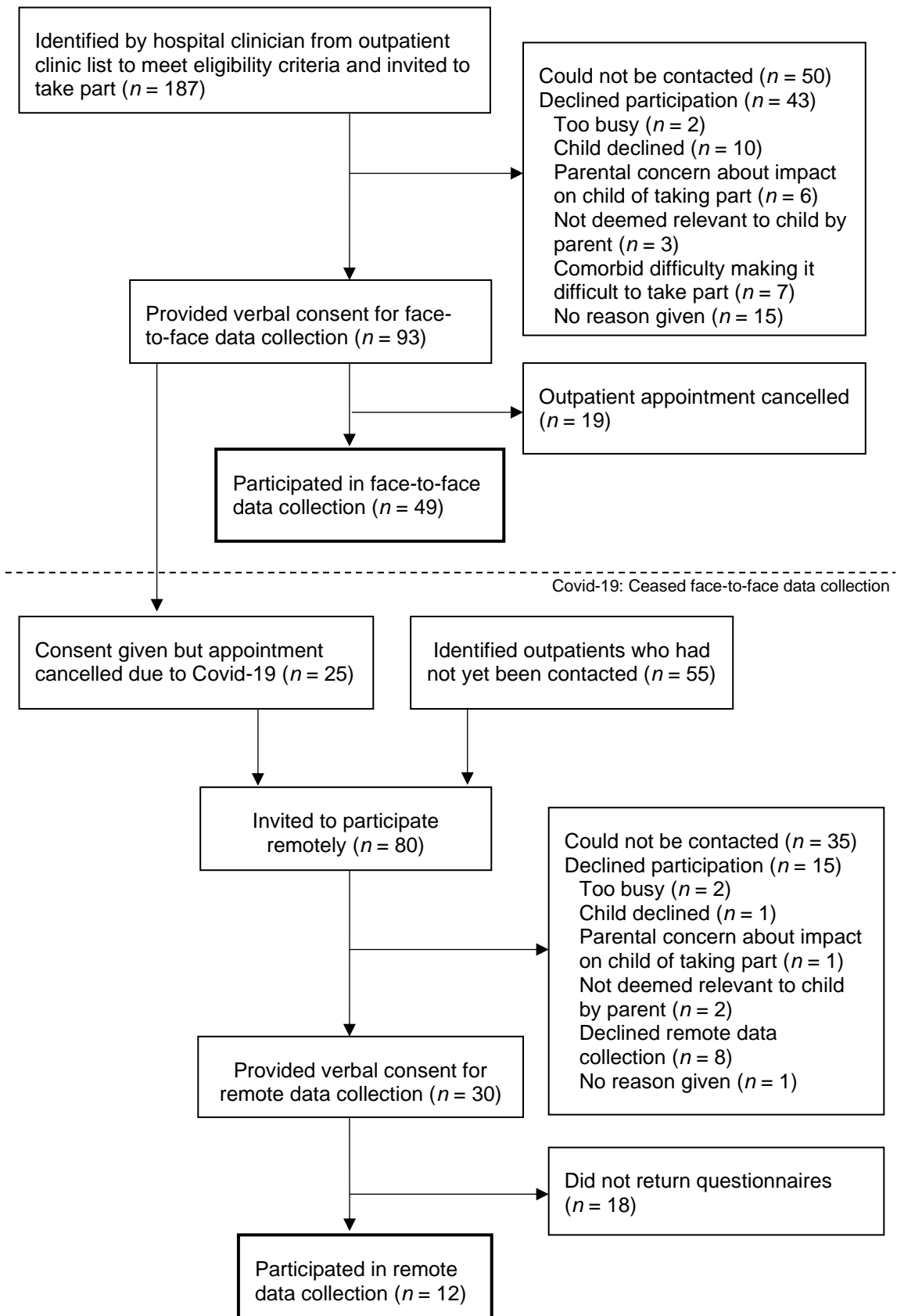
3.3. Measures

3.3.1. Patient and Public Involvement

A focus group from the paediatric hospital's Young People's Committee (YPC) consisting of children aged 8–14 years reviewed the measures to be completed by the child participants prior to data collection, to ensure that the language used in the questionnaires was understandable to children in the same age range. The YPC consisted of current and former patients, who are consulted on a range of hospital affairs including staff recruitment, research and fundraising. The unanimous group feedback indicated that the questionnaires were readable and understandable, and

Figure 1

Flowchart of Stages of Recruitment and Data Collection



the instructions easy to follow. They sought clarification on the term “health condition”, which was changed to “medical condition”.

3.3.2. *Child Stigma Scale*

This 8-item scale was used to measure physical health stigma in children (Austin et al., 2004, see Appendix B). Although initially developed for 9-14 year olds with epilepsy, this scale has since been adapted and used with children who have other physical health conditions (e.g. Gamwell et al., 2018; Rolston et al., 2015; Wakefield et al., 2017) and child mental health populations (Kaushik et al., 2017). The developers reported it to have good internal consistency reliability ($\alpha = .81$) and construct validity based on correlations with mental health and self-efficacy measures (Austin et al., 2004). The Child Stigma Scale asks children to rate each item, for example: “*How often do you feel different from other kids because you have a medical condition?*”, on a 5-point scale from 1 (“*Never*”) to 5 (“*Very often*”). The total score is calculated by summing the items and dividing by the total number of eight items. The total score ranges from 1-5, where higher score reflects greater perceptions of stigma.

3.3.3. *Parent Stigma Scale*

This 5-item scale was used to measure parental stigma of their child’s health condition (Austin et al., 2004, see Appendix B). The Parent Stigma Scale was developed together with the Child Stigma Scale, and was deemed to have good internal consistency ($\alpha = .79$) and construct validity based on correlations with related constructs of parental worry and negative impact of epilepsy on family life (Austin et al., 2004). Parents were asked to rate items, for example: ‘*My child always has to prove him/ herself because of their medical condition*’, on a 5-point scale ranging from 1 (“*Strongly disagree*”) to 5 (“*Strongly agree*”). The total score is calculated by summing the items and dividing by the total number of five items. The total score ranges from 1-5, where higher scores reflect greater parental perceptions of stigma.

3.3.4. *Child Attitude Towards Illness Scale (CATIS)*

This 13-item scale is a measure of children's illness perceptions (Austin et al., 1993, see Appendix B). It was originally developed for children aged 8-12 years, but has since been validated up to the age of 17 years and has been deemed to have good internal consistency ($\alpha = .89$) and test-retest reliability ($r = .77$) (Heimlich et al., 2000). The CATIS asks children to rate each item, for example: "*How good or bad do you feel it is that you have a medical condition?*", on a 5-point scale ranging from 1–5 (with varying responses, see Appendix B). The total score is calculated by summing the items and dividing by the total number of 13 items. The total score ranges from 1-5, where higher scores reflect more positive illness perceptions (some items are reverse-coded). As this measure pertains to illness attitudes in the context of lived experience of having a health condition, no adaptation was made to create an equivalent parental measure of illness perceptions.

3.3.5. *Secrecy Scale (Child and Parent Versions)*

This 7-item scale was used to measure attitudes towards concealment of the chronic health condition by children and their parents (see Appendix A and B). It was adapted from the Paediatric Self-Stigmatisation Scale (Kaushik et al., 2017); a measure of concealment of mental health problems in children aged 8-12 years. The original measure was found to have good internal consistency ($\alpha = .79$) in this population. Here, it has been adapted by changing the phrase "*difficult feelings and behaviour*" to "*medical condition*". The Secrecy Scale asks children to rate each item, for example: "*I often feel the need to hide the fact that I have a health condition*", from 0 ("*I disagree a lot*") to 4 ("*I agree a lot*"). For parents, the same items were adapted so that '*I*' was replaced with '*my child*', for example: '*I often feel the need to hide the fact that my child has a medical condition*'. Scores range from 7–28, with higher scores representing attitudes supporting higher levels of concealment of the health condition.

3.3.6. *Strengths and Difficulties Questionnaire (SDQ)*

The Internalising Score of the SDQ (consisting of the 'emotional symptoms' and 'peer relationship problems' subscales) was used as a measure of children's psychosocial difficulties (Goodman, 2001, see Appendix A). The SDQ was designed for use with 4–17 year olds, and was reported by the developers to have satisfactory internal consistency ($\alpha = .73$), satisfactory inter-rater agreement, and moderate to high test-retest reliability ($\alpha = .51-.80$) (Goodman, 2001). The Internalising Score has been recommended for use in non-clinical samples and found to have superior construct validity compared with using the total score (Goodman et al., 2010). In this study, the parent-rated version was used to reduce the questionnaire burden in children. Parents were asked to rate each item about their child, for example: "*Many worries, often seems worried*", on a 3-point scale, ranging from 0 ("*Not true*") to 2 ("*Certainly true*"). The SDQ Internalising Score ranges from 0-20, which higher scores indicating greater emotional and social difficulties.

3.3.7. *Psychometric Properties of the Measures in this Sample*

The internal consistencies of the measures used for the sample of children and parents in this study were calculated. Good internal consistency was found for the CATIS ($\alpha = .871$), the Child Stigma Scale ($\alpha = .887$), the Child Secrecy Scale ($\alpha = .858$), the Parent Secrecy Scale ($\alpha = .802$). Acceptable internal consistency was found for the Parent Stigma Scale ($\alpha = .729$) and the SDQ Internalising score ($\alpha = .755$).

3.4. Procedure

Potential participants were identified by a member of the care team from outpatient clinic lists according to the inclusion criteria. Eligible children and their parents were sent a study invitation pack four weeks prior to the child's next scheduled outpatient hospital appointment, including a letter of invitation (Appendix C), a child participant information sheet (Appendix D), and a parental participant information sheet

(Appendix E). A follow-up telephone call was made two weeks later to answer any questions about the study, to determine if inclusion/exclusion criteria were met, and to arrange a meeting to confirm written consent and to collect data with the parent/guardian if they were willing to participate.

Children and parents who agreed to participate were met by researchers at the child's outpatient clinic appointment, where the researcher conducted the formal consenting procedure. Signed child assent forms (Appendix F), parental consent forms on behalf of the child (Appendix G), and parental consent forms (Appendix H) were collected. Researchers supported participants in completing the measures either before or after the child's appointment, depending on participant preferences. This generally took 10-30 minutes. Researchers debriefed participants, checked if they were distressed by completing the questionnaires, and offered support/signposting as required. The same procedure was followed for remote data collection, but via telephone, email or video call (depending on participant preference), whereby written consent was collected and participants were offered support when completing the questionnaires as required. All participants were asked if they would like to be informed of the results at the end of the study. Those who did were asked for their contact details.

3.5. Ethical Considerations

This study received ethical approval from the Health Research Authority (Integrated Research Approval System project ID number: 256531, see Appendix I) and approval from the Clinical Research Adoptions Committee at Great Ormond Street Hospital (Research and Development number: 19SH02, see Appendix I).

3.6. Power Analysis

Power analysis was completed using a number of studies which used correlational analysis to investigate similar constructs or measures in similar populations (Austin et al., 2004; Benson, 2016; Carlton-Ford et al., 1997; Corrigan et al., 2016; LeBovidge

et al., 2005; Quinn et al., 2014). These studies found moderate to large effect sizes (Cohen, 1992). Power calculations were carried out using G*Power 3.1 software (Faul et al., 2007) based on information from the literature and the planned statistical analyses, with the alpha value specified as 5% and the desired power as 80%. The analysis estimated a required sample size of 63 child-parent pairs.

3.7. Statistical Analyses

The data was analysed using SPSS Version 26. The z-scores were calculated on all six measures to identify any outliers (where $z \geq 3.29$), and none were identified. Tests of normality indicated that data was normally distributed in most measures, but the SDQ Internalising Score and Parent Secrecy Scale were positively skewed.

One-sample t-tests were used to compare mean scores with previous studies. Preliminary analyses were used to determine group differences based on demographic factors or children's clinical profiles for each measure. Independent samples t-tests and one-way ANOVAs were used to compare group means on the normally distributed measures, and Mann-Whitney U and Kruskal-Wallis tests were used to compare group medians on the SDQ Internalising Score and Parent Secrecy Scale. Where several covariates were identified, correlational analyses and guidance regarding the number of predictors, sample size and adequate statistical power were used to determine which covariates to control for in subsequent planned regression analyses (Field, 2013; Miles et al., 2001).

To account for some measures not being normally distributed, both Pearson's and Spearman's Rho bivariate correlations were conducted. As there was a difference in which associations were significant, Spearman's Rho was used and differences in significance were reported, see Appendix L. Following preliminary analyses to ensure no violation of the assumptions of normality, homoscedasticity, linearity, and multicollinearity, several multiple hierarchical regression analyses were carried out. First,

these analyses were carried out to determine the predictors of child concealment and of parental concealment. Next, several independent hierarchical multiple regressions were conducted to determine predictors of children's psychosocial difficulties. This was due to the large number of possible predictor variables in a small sample size and the correlations between some predictors. Significant predictors were then added together in a final hierarchical multiple regression. Although all assumptions were met, it should be noted that the regression analyses for children's psychosocial difficulties all had slight positive skew, meaning that generalisation of the model is not advised.

A principal component analysis (PCA) was conducted on the Child Stigma Scale, to determine whether it measured more than one type of stigma. Key analyses were repeated with the resultant two components but as there was no difference in the significance of results, they were not used as distinct constructs in the main analyses.

3.8. Joint Thesis Declaration

This is a joint thesis conducted together with Jemma Ambrose, who was investigating the stigma experiences of children with visible and less visible physical health conditions (Ambrose, 2020). See Appendix J for further details.

4. Results

4.1. Sample Characteristics

A total of 61 child-parent pairs took part. Children were aged between 8-14 years ($M = 12.0$, $SD = .5$), and parents/caregivers were aged between 32-70 years ($M = 42.8$, $SD = 8.0$), and consisted of 50 mothers (82%), 9 fathers (14.8%), 1 grandmother (1.6%) and 1 grandfather (1.6%). Participants' full demographic characteristics and children's clinical profiles are presented in Table 1.

4.2. Child Stigma Scale, Parent Stigma Scale, CATIS, Child Secrecy Scale, Parent Secrecy Scale, and SDQ Internalising Scores

Table 2 presents the means and standard deviations for each measure in this sample, together with previous mean scores from comparative studies. On the Child Stigma Scale, the mean response was between “sometimes” and “often”, indicating that children perceived others to be stigmatising towards them ($M = 2.40$, $SD = .94$). The mean response on the CATIS ($M = 3.22$, $SD = .71$) indicated neutral to positive illness perceptions. On the Child Secrecy Scale, the mean response was between “disagree” and “agree” ($M = 2.42$, $SD = .68$), indicating a neutral stance towards concealment.

Table 1

Sociodemographic Characteristics and Clinical Profiles of Participants

Characteristics	n	%
Child gender		
Female	31	50.8
Male	30	49.2
Child School Year		
Year 3-4	7	11.5
Year 5-6	16	26.2
Year 7-8	18	29.5
Year 9-10	20	32.8
Child Ethnicity		
White British/ White Other	46	75.4
Asian/ Asian British	7	11.5
Black/ Black British	4	6.6
Other / Prefer not to say	4	6.6
Clinic		
Dermatology	35	57.4
Urology	26	42.6
Medical condition		
Eczema	19	31.1
Epidermolysis bullosa	9	14.8
Psoriasis	6	9.8
Bladder/ incontinence	12	19.7
Kidney problems	15	24.6
Comorbidities ^a	18	29.5
Age of onset		
Present at birth	24	39.3

Less than 5 years ago	23	37.7
5 years or older	14	23.0
Hospital attendance		
1-2 times a year	30	49.2
3-10 times a year	25	41.0
More than 10 times a year	6	9.8
Caregiver gender		
Female	51	83.6
Male	10	16.4
Caregiver ethnicity		
White British/ White Other	46	75.4
Asian/ Asian British	8	13.1
Black/ Black British	4	6.6
Other / Prefer not to say	3	4.9

^a Reflects the number and percentage of participants answering “yes” to this question

Table 2

Means and Standard Deviations in the Current Sample and Previous Studies for Child and Parent-Rated Measures of Stigma, Concealment, Illness Perceptions, and Children’s Psychosocial Difficulties

Measure	Current sample		Austin et al. (2004)		Austin et al. (1993)		Kaushik et al. (2017)		de la Cruz et al. (2018)	
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>
Child Stigma Scale	2.40	.94	2.42	.68	-	-	-	-	-	-
Parent Stigma Scale	2.45	.77	2.58	.81	-	-	-	-	-	-
CATIS	3.22	.71	-	-	3.2	.5	-	-	-	-
Child Secrecy Scale	2.42	.68	-	-	-	-	2.70	.69	-	-
Parent Secrecy Scale	1.97	.51	-	-	-	-	-	-	-	-
SDQ Emotional Subscale	4.00	2.60	-	-	-	-	-	-	4.7	2.8
SDQ Peer Problems Subscale	2.29	2.25	-	-	-	-	-	-	3.6	2.4

The mean score for children's psychosocial difficulties (SDQ Internalising Score) was 6.30 ($SD = 4.04$), where higher scores represent greater psychosocial difficulties (possible range of 0-20). The mean parent stigma (Parent Stigma Scale) score ($M = 2.45$, $SD = .77$) indicated a neutral stance, corresponding with an answer of "neither" agreeing or disagreeing with the statements. The mean parent concealment (Parent Secrecy Scale) score ($M = 1.97$, $SD = .51$) corresponded to an answer of "disagree", indicating a negative stance towards concealment.

One sample t-tests revealed no significant differences in scores compared to previous samples of children with epilepsy on the Child Stigma Scale, the Parent Stigma Scale, or the CATIS (Austin et al., 1993; Austin et al., 2004). Although this sample had significantly lower Child Secrecy Scale scores than the original sample ($t(60) = -3.16$, $p < .00$), the original study involved children with mental health rather than physical health conditions (Kaushik et al., 2017). As the Parent Secrecy Scale was adapted for this study, there are no comparative sample scores. The SDQ subscales comprising the Internalising Scores were significantly lower than those reported in a large UK paediatric sample (de la Cruz et al., 2018), in both the emotional ($t(60) = -2.10$, $p < 0.04$) and the peer problems subscale ($t(60) = -4.54$, $p < 0.00$), suggesting this sample had lower levels of psychosocial difficulties.

4.3. Preliminary Analyses of Demographic and Clinical Factors

Group differences based on demographic factors and children's clinical profiles on each measure was evaluated, see Appendix K for the full results. The significant results for child concealment, parent concealment, and children's psychosocial difficulties are presented below, as the preliminary analyses used to decide which covariates to control for in regression analyses. There were no significant differences between groups in any other demographic or clinical factors for child stigma, or based on whether data collection was conducted face-to-face or remotely.

4.3.1. Child Concealment and Group Differences in Demographic and Clinical Factors

Child concealment scores differed significantly between medical conditions ($F(4,56) = 3.090, p = .023$). Children with psoriasis had the highest mean score ($M = 2.98, SD = .51$), followed by bladder-related conditions ($M = 2.71, SD = .58$), epidermolysis bullosa ($M = 2.54, SD = .83$), eczema ($M = 2.27, SD = .65$), and kidney-related conditions ($M = 2.09, SD = .68$). Bonferroni post-hoc analysis revealed no significant differences between any of the groups ($p > .05$). There were no significant differences between groups in any other demographic or clinical factors, and medical condition was therefore identified as a covariate for regression analyses where child concealment was the dependent variable.

4.3.2. Parent Concealment and Group Differences in Demographic and Clinical Factors

Parent concealment scores differed significantly between groups based on the child's gender ($U = 309.000, z = -2.264, p = .024$), where parents of male children ($Md = 2.07, N = 30$) reported higher levels of concealment than parents of female children ($Md = 1.86, n = 21$). Parent concealment scores also differed based on medical speciality ($U = 266.500, z = -2.811, p = .005$), where parents scored higher median concealment scores in the urology clinics ($Md = 2.14, n = 27$) than in the dermatology clinics ($Md = 1.86, n = 34$). Significant differences were also found between medical conditions ($H(4) = 14.036, p = .007$), where pairwise comparisons with a Bonferroni correction revealed significant parental concealment in both children with kidney conditions ($Md = 2.00, n = 15$) and bladder conditions ($Md = 2.14, n = 12$) than in children with epidermolysis bullosa ($Md = 1.43, n = 9$), but there were no other significant group differences between children with eczema ($Md = 2.00, n = 19$) or psoriasis ($Md = 1.72, n = 6$). There were no other significant differences between groups in any other demographic or clinical factors for parent concealment.

Both children's gender ($r_s = .292, p = .022$) and medical speciality ($r_s = .363, p = .004$) were identified as covariates to include in regression analyses due to significant associations with higher levels of parental concealment, whereas medical conditions was not ($r_s = .190, p = .14$).

4.3.3. Children's Psychosocial Difficulties, and Group Differences in Demographic and Clinical Factors

For children's psychosocial difficulties, a significant difference in scores based on parent gender was revealed ($U = 149.000, z = -2.075, p = .038$), where female parents had higher median scores ($Md = 2.00, n = 51$) than male parents ($Md = 1.93, n = 10$). Scores of children with comorbidities ($Md = 8.00, n = 18$) were significantly higher than those without ($Md = 5.00, n = 43$) ($U = 255.000, z = -2.098, p = .036$). Children's psychosocial difficulties also differed between groups based on the frequency of hospital attendance ($H(2) = 8.489, p = .014$). Pairwise comparisons with a Bonferroni correction revealed that the only significant difference was between children who attended hospital "1-2 times per year" ($Md = 4.00, n = 30$) and "10+ times per year" ($Md = 8.50, n = 6$), but not with those attending "3-10" times per year ($Md = 7.00, n = 25$). There were no other significant differences between groups in any other demographic or clinical factors for children's psychosocial difficulties.

Frequency of hospital attendance showed the strongest correlation with children's psychosocial difficulties ($r_s = .375, p = .003$), and was therefore prioritised as a covariate in future regression analyses where psychosocial difficulties was the dependent variable. Male parent gender ($r_s = -.268, p = .037$) and children having a comorbid condition ($r_s = -.271, p = .035$) were also associated with higher levels of children's psychosocial difficulties, but with a small effect size.

4.4. The Relationship Between Child and Parental factors of Stigma, Concealment and Illness perceptions

4.4.1. Relationship between Child Stigma, Child Concealment and Illness Perceptions

Table 3 displays the bivariate correlations from a Spearman's Rho analysis between all child and parent variables of stigma, concealment and illness perceptions. Higher levels of child stigma were associated with higher levels of child concealment ($r_s = .566, p < .001$) and more negative illness perceptions ($r_s = -.566, p < .001$), both with a large effect size. However, child concealment and child illness perceptions were not significantly correlated ($p > .05$).

Table 3

Spearman's Rho Correlations between Measures of Stigma, Concealment, and Illness Perceptions

Measure	1	2	3	4	5
1.Child Stigma	–				
2.Child Concealment	.566**	–			
3.Child Illness Perceptions	-.562**	-.196	–		
4.Parent Stigma	.199	.127	-.195	–	
5.Parent Concealment	-.013	-.064	.079	.331*	–

Note. $N = 61$ on all measures. * $p < .05$. ** $p < .01$

4.4.2. Relationship between Child and Parental Factors of Stigma and Concealment

Child stigma was not significantly correlated with either parent stigma or parent concealment ($p > .05$). Child concealment was also not significantly correlated with either parent stigma or parent concealment ($p > .05$). Higher levels of parent stigma were associated with higher levels of parent concealment ($r_s = .331, p = .009$), with a moderate effect size.

4.5. Child and Parental Predictors of Concealment

Child stigma emerged as an independent predictor of child concealment even when controlling for the child's medical condition, but parent stigma and parent concealment did not. The full model was statistically significant ($R^2 = .379$, $F(4,56) = 8.549$, $p > .001$, adjusted $R^2 = .335$), and the addition of medical condition as a control in Model 2 added 1% of variance to the model. See Table 4 for full details. This model showed slight heteroscedasticity, meaning generalisation of the model is not advised.

Parent stigma emerged as an independent predictor of parent concealment when controlling for child gender and the medical specialty (urology or dermatology), see Table 5 for full details. The full model was statistically significant ($R^2 = .330$, $F(3,57) =$

Table 4

Hierarchical Multiple Regression for Child Concealment

Variable	Child Concealment			
	Model 1		Model 2	
	B	β	B	β
Constant	1.43**		1.31**	
Child Stigma	.44**	.61	.45**	.62
Parent Concealment	-.04	-.03	-.10	-.07
Parent Stigma	.01	.10	.04	.04
Medical condition			.05	.11
R^2	.37		.38	
F	11.10**		8.55**	
ΔR^2	.37		.01	
ΔF	11.10**		.93	

Note. $N = 61$. * $p < .05$. ** $p < .01$

Table 5

Hierarchical Multiple Regression for Parental Concealment

Variable	Parental Concealment			
	Model 1		Model 2	
	B	β	B	β
Constant	1.40**		.56*	
Parent Stigma	.23**	.35	.24**	.36
Child Gender			.19	.12
Medical Specialty			.38**	.37
R^2	.12		.33	
F	8.05**		9.37**	

Note. $N = 61$. B = unstandardised regression coefficient; β = standardised coefficient; R^2 = Coefficient of determination. * $p < .05$. ** $p < .01$.

9.373, $p < .001$, adjusted $R^2 = .210$). Adding child gender and medical specialty accounted for an additional 21% of the variability in parental concealment, and medical specialty also emerged as an independent predictor.

4.6. Psychosocial Difficulties, and Child and Parental factors of Stigma, Concealment and Illness Perceptions

4.6.1. The Relationship between Children's Psychosocial Difficulties, Stigma, Concealment and Illness Perceptions

In a Spearman's Rho bivariate correlational analysis, higher levels of children's psychosocial difficulties were associated with higher levels of child stigma ($r_s = .379$, $p = .003$) and parent stigma ($r_s = .371$, $p = .003$), both with a medium effect size, and with more negative child illness perceptions ($r_s = -.284$, $p = .027$), with a small effect size. Neither child or parent concealment significantly correlated with children's psychosocial difficulties ($p < .05$), see Table 6 for full details. Appendix L presents the Pearsons correlational analysis results, which found similar associations with greater effect sizes for child stigma ($r = .459$, $p < .001$), parent stigma ($r = .386$, $p = .002$) and

child illness perceptions ($r = -.397, p = .002$), and additionally found a significant association with child concealment ($r = .275, p = .032$), with a small effect size.

Table 6

Spearman's Rho Correlations of Children's Psychosocial Difficulties with Child and Parent Factors Stigma, Concealment and Illness Perceptions

Measure	Child Psychosocial Difficulties
Child Stigma	.379**
Child Concealment	.236
Child Illness Perceptions	-.284*
Parent Stigma	.371**
Parent Concealment	.007
Child Psychosocial Difficulties	–

Note. $N = 61$ on all measures. * $p < .05$. ** $p < .01$

4.6.2. Predictors of Child Psychosocial Difficulties

4.6.2.1. Child Psychosocial Difficulties and Child Stigma

When entered into the model independently, child stigma accounted for 21% of the variance in children's psychosocial difficulties ($F(59,60) = 15.718, p < .001$). After controlling for the frequency of hospital attendance, the full model accounted for 32% of the variance in children's psychosocial difficulties ($F(2,58) = 13.535, p < .001, R^2$ change = .108), see Table 7 for full details. Both child stigma and frequency of hospital attendance were significant predictors of children's psychosocial difficulties.

4.6.2.2. Child Psychosocial Difficulties and Child Illness Perceptions

Child illness perceptions accounted for 16% of the variance in children's psychosocial difficulties when entered into the model independently ($F(1,59) = 11.025, p = .002$). The full model including frequency of hospital attendance accounted for 22% of the variance ($F(2,58) = 9.505, p < .001, R^2$ change = .089), with both child illness perceptions and hospital attendance emerging as statistically significant. See Table 7

for full details. It should be noted that there was slight heteroscedasticity, meaning generalisation of the models is not advised.

4.6.2.3. Child Psychosocial Difficulties and Child Concealment

Child concealment accounted for 8% of the variance in children's psychosocial difficulties when entered independently ($F(1,59) = 4.840, p = .032$). The full model including frequency of hospital attendance accounted for 20% of the variance ($F(2,58) = 7.530, p = .001, R^2 \text{ change} = .126$), where child concealment did not emerge as a significant predictor hospital attendance had been controlled for. See Table 7 for full details.

Table 7

A Series of Multiple Hierarchical Regressions with Child Stigma, Child Illness Perceptions, Child Concealment, and Parent Stigmas as Predictors of Children's Psychosocial Difficulties, Controlling for Frequency of Hospital Attendance

Variable	Child Psychosocial Difficulties			
	Model 1		Model 2	
	<i>B</i>	β	<i>B</i>	β
1. Child Stigma				
Constant	1.56		-1.18	
Child Stigma	1.98**	.46	1.77**	.41
Frequency of hospital attendance			2.02**	.33
R^2	.21		.32	
<i>F</i>	15.72**		13.54**	
2. Child Illness Perceptions				
Constant	13.53**		9.06	
Illness Perceptions	-2.25**	-.397	-1.80**	-.32
Frequency of hospital attendance			1.88*	.31
R^2	.16		.25	
<i>F</i>	11.03**		9.51**	
3. Child Concealment				
Constant	2.35		-.42	

Child Concealment	1.63*	.28	1.33	.22
Frequency of hospital attendance			2.18**	.36
R^2	.08		.20	
F	4.84*		7.35**	
4. Parent Stigma				
Constant	1.35		-.82	
Parent Stigma	2.02**	.39	1.64*	.31
Frequency of hospital attendance			1.94**	.32
R^2	.15		.25	
F	10.33**		9.45**	

Note. $N = 61$. B = unstandardised regression coefficient; β = standardised coefficient; R^2 = Coefficient of determination. * $p < .05$. ** $p < .01$.

4.6.2.4. Child Psychosocial Difficulties and Parent Stigma

Parent stigma accounted for 15% of the variance in children's psychosocial difficulties when entered into the model independently ($F(59,60) = 10.334, p = .002$). After controlling for the frequency of hospital attendance, the full model accounted for 25% of the variance in children's psychosocial difficulties ($F(2,58) = 9.438, p < .001, R^2$ change = .096), see Table 7 for full details. Both parent stigma and frequency of hospital admission were significant predictors of children's psychosocial difficulties.

4.6.2.5. Child Psychosocial Difficulties, Child Stigma, Parent Stigma and Child Illness Attitudes

A hierarchical multiple regression model was run to determine the relevant contribution of each predictor for children's psychosocial difficulties, while controlling for frequency of hospital admission. See Table 8 for full details. This model showed slight heteroscedasticity, meaning generalisation of the model is not advised. Child stigma and parent stigma explained 28.9% of the variance in Model 1. Adding the frequency of hospital attendance as a control in Model 2 explained an additional 7.5%

of the variance. The full model (Model 2) of child stigma, parent stigma and frequency of hospital admission was statistically significant ($R^2 = .373$, $F(3,57) = 11.286$, $p < .001$). Each predictor also emerged as independently significant, with child stigma recording the highest beta value. As adding child illness perceptions added for 0% additional variability ($p > .05$), this was not included in the final model.

Table 8

Multiple Hierarchical Regression Predicting Child Psychosocial Difficulties from Child Stigma and Parent Stigma, Controlling for Frequency of Hospital Attendance

Variable	Child Psychosocial Difficulties			
	Model 1		Model 2	
	<i>B</i>	β	<i>B</i>	β
Constant	-1.66		-3.36	
Child Stigma	1.70**	.40	1.58**	.37
Parent Stigma	1.59**	.30	1.23*	.24
Frequency of hospital attendance			1.71**	.28
R^2	.30		.34	
<i>F</i>	12.32**		11.29**	

Note. $N = 61$. *B* = unstandardised regression coefficient; β = standardised coefficient; R^2 = Coefficient of determination. * $p < .05$. ** $p < .01$.

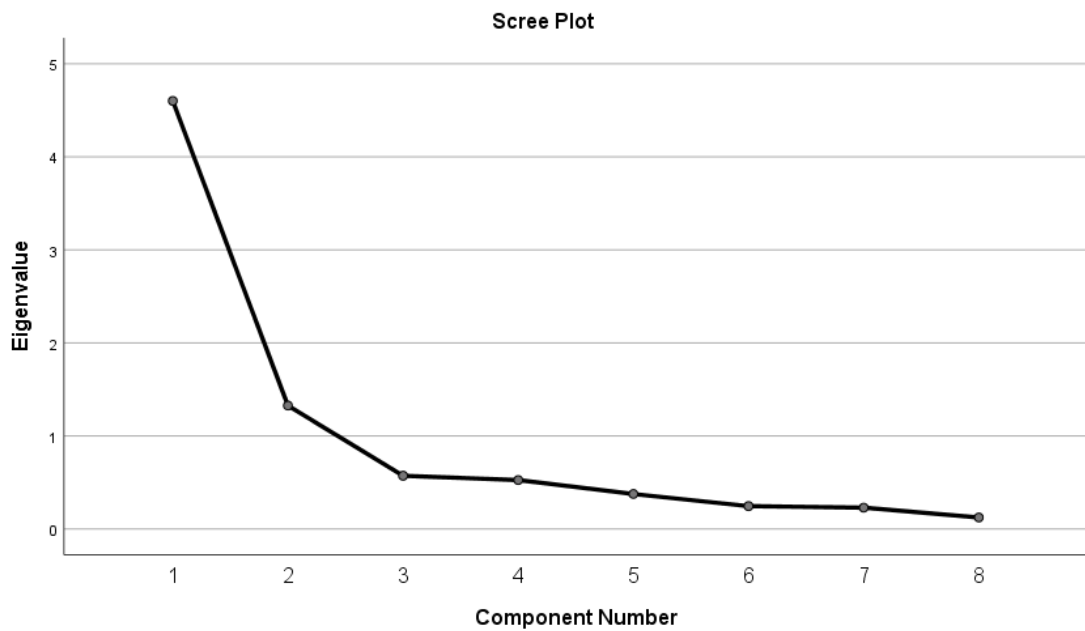
4.7. Principal Component Analysis of the Child Stigma Scale

The eight items of the Child Stigma Scale were subjected to PCA. The following tests confirmed that the data was suitable for PCA: the correlation matrix showed multiple coefficients of .3 or higher, the Kaiser-Meyer-Olkin (Kaiser, 1960) was higher than .6 (KMO = .799), and Bartlett's Test of Sphericity (Bartlett, 1954) supported the factorability of the correlation matrix ($\chi^2(28) = 301.505$, $p < .001$).

Two components had eigenvalue factors exceeding 1, explaining 57.5% and 15.6% of the variance respectively. Although the scree plot showed inflexions at both components 1 and 2 (see Figure 2), the Cattell (1966) scree test was used to inform

Figure 2

Scree plot of Components of the Child Stigma Scale Items



the decision to retain two components for further investigation. The two components explained a total of 74.1% of the variance, and Table 9 shows the factor loadings after an oblimin rotation. The item clustering suggests that component 1 represents 'perceived stigma' and component 2 represents 'self stigma'. Question 1 ("How often do you feel different from other kids because you have a medical condition?") loaded onto both components, suggesting it was very broad, and was therefore removed.

When the associations between all factors were compared with the new components of Child Self-Stigma ($M = 1.95$, $SD = .97$) and Child Perceived Stigma ($M = 2.83$, $SD = 1.26$), there was no difference in which other variables were significantly correlated with these two components (see Table 10) compared with when Child Stigma was entered as one construct (see Table 3).

Table 9

Results from a Principal Component Analysis of the Child Stigma Scale

Child Stigma Scale Item	Pattern coefficients		Structure coefficients	
	1	2	1	2
Factor 1: Perceived stigma				
3. How often do you feel other children are uncomfortable with you because of your medical condition?	.960	-.140	.908	.423
4. How often do you feel people may not want to be friends with you if they know you have a medical condition?	.903	.011	.896	.299
2. How often do you feel people may not like you if they know you have a medical condition?	.836	.083	.873	.465
5. How often do you feel people would not want to go out with you or ask you to parties if they know you have a medical condition?	.772	.033	.787	.386
Factor 2: Self-stigma				
8. How often do you try to avoid talking to other people about your medical condition?	-.079	.915	.364	.886
7. How often do you keep your medical condition a secret from other kids?	-.052	.910	.339	.879
6. How often do you feel embarrassed about your medical condition?	.337	.659	.638	.813

Note. Factor loadings above .40 are in bold. Eigenvalues for Factor 1 = 4.601, Factor 2 = 1.327.

Table 10

Spearman's Rho Correlations between Child Self-Stigma, Child Perceived Stigma, Concealment, Illness Perceptions, and Child Psychosocial Difficulties

Measure	1	2	3	4	5	6	7
1.Child Self-stigma	–						
2.Child Perceived Stigma	.541**	–					
3.Child Concealment	.401**	.612**	–				
4.Child Illness Perceptions	-.569**	-.385**	-.196	–			
5.Parent Stigma	.244	.125	.127	-.195	–		
6.Parent Concealment	-.005	-.002	-.064	.079	.331**	–	
7. Child Psychosocial Difficulties	.334**	.345**	.236	-.284*	.371**	.007	–

Note. $N = 61$ on all measures. * $p < .05$. ** $p < .01$

However, self-stigma and perceived stigma in children had different effect sizes in their correlations with other variables. Both were positively correlated with child concealment, but for self-stigma the effect size was moderate ($r_s = .401$, $p = .001$) whereas for perceived stigma the effect size was large ($r_s = .612$, $p < .001$). Whilst both were negatively correlated with child illness perceptions, for child self-stigma the effect size was large ($r_s = -.569$, $p < .001$), whereas for the child perceived stigma the effect size was moderate ($r_s = -.385$, $p = .002$). No parent and child factors revealed any significant correlations with each other ($p > .05$).

5. Discussion

The overall purpose of this study was to understand the associations between stigma, concealment, illness perceptions, and psychosocial difficulties in children with physical health conditions and their parents. Additional aims were to identify which child and parent-rated factors predicted: 1) concealment, and 2) children's psychosocial difficulties.

5.1. The Relationships between Child and Parental Factors of Stigma, Concealment and Illness Perceptions

No associations were found between any child-rated and parent-rated factors in this study. This is in contrast with previous studies in children with epilepsy which supported the idea of 'stigma coaching' (Austin et al., 2004; Benson et al., 2016; Ryu et al., 2015), although equivalent mean scores for child and parent stigma were found in this study compared with one of these previous studies (Austin et al., 2004). However, the effect sizes of previously documented associations between child and parental factors were not large ($r \leq .31$). In children with mental health conditions, parental stigma was also not associated with child-rated stigma or concealment (Kaushik et al., 2017), and Corrigan et al. (2007) cite a series of studies where no associations were found between children and parents based on public stigma about race. With mixed results in the wider literature, the findings in this study suggest that child and parental factors should not be assumed to be associated with each other.

A possible explanation for this finding is that children are more influenced by external factors such as public stigma. Peer acceptance is seen as increasingly important through middle childhood and adolescence (Pittman et al., 2007), and peer attitudes towards physical health conditions may therefore be prioritised by children. Public stigma in school children has been documented across a number of health conditions, with young people reporting that they would be less likely to be friends with peers with health conditions, would expect them to be bullied, or to have difficulties with dating (Austin et al., 2002; Gupta et al., 2018; Masnari et al., 2013b). If parents are not influencing children's stigma perceptions, then addressing public stigma in children's peer groups may be a more useful focus for intervention.

There were a number of interesting findings regarding child stigma and its relationship with other factors. Firstly, both concealment and illness perceptions were associated with stigma in children. This replicates findings in the adult stigma literature, and

highlights the relevance of including concealment and illness perceptions in stigma research (Quinn et al., 2014). These associations are discussed further in the sections below, in the context of concealment and psychosocial difficulties. Second, PCA revealed two possible components within the Child Stigma Scale, which were labelled as 'self-stigma' and 'perceived stigma'. Although the significance of correlations did not differ when using these sub-scales compared with the full Child Stigma Scale, there were differences in effect sizes: perceived stigma was more strongly correlated with concealment, whereas self-stigma was more strongly correlated with illness perceptions. The development of separate validated measures of self-stigma and perceived stigma could investigate whether these preliminary findings are replicable. Finally, child stigma scores did not significantly differ between any groups based on demographic or clinical factors, suggesting that factors like age, gender, ethnicity and the nature of a child's medical condition did not affect children's stigma experiences in this study.

5.2. Stigma and Concealment in Children and Parents

Stigma was found to be associated with the concealment of children's health conditions in both children and parents. In the context of this association with stigma, concealment can be understood as children's attempts to conform to 'normality' by hiding their health condition as a part of their identity. The decision about whether to disclose or conceal a child's medical condition has been consistently reported as difficult: although concealment has been cited as a protective strategy by children and parents, it reduces opportunities for both informal and formal support (Chavira et al., 2017; Moola et al., 2011; Pachankis, 2007). The findings of this study build on previous associations between stigma and concealment in children with epilepsy and their parents (Benson et al., 2016), but also introduces the idea that the nature of a child's medical condition may affect decisions about concealment. Medical speciality (dermatology or urology) emerged as an independent predictor for parental

concealment, with higher concealment scores in urological conditions. Although dermatological conditions are more likely to be visible and might therefore have been expected to have higher concealment scores (Kaushansky et al., 2017), a reluctance of parents to bring up their children's bladder-related issues to healthcare professionals due to shame has previously been documented (Cederblad et al., 2014), and may therefore account for higher concealment by parents in children with urological conditions. Child concealment differed based on their medical condition, with the highest concealment scores reported by children with psoriasis. Future research with a mixed methods approach could further investigate how concealment practices might vary across medical conditions in children and their parents, and the reasons for this.

The association between stigma and concealment may also represent a barrier to change: the act of concealment could reduce the opportunities for stigma to be challenged, where both stigma and concealment are inadvertently perpetuated. This has previously been evidenced in a qualitative study across a number of chronic health conditions, which found there were fears of rejection and isolation, but few examples of when these had been realised (Kaushansky et al., 2017). The finding that stigma and concealment are associated has possible implications. Interventions targeting stigma and disclosure decisions in mental health have found improvements in stigma-related stress, disclosure-related distress and help-seeking (Mulfinger et al., 2018): adaptations of such interventions for children with physical health conditions could be explored in future research.

5.3. Children's Psychosocial Difficulties and Child and Parental Factors of Stigma, Concealment and Illness Attitudes

Higher levels of child stigma were associated with higher levels of children's psychosocial difficulties. This builds on similar findings in children with other physical health conditions (Austin et al., 2004; Taft et al., 2009), to suggest that stigma should

be considered a risk factor for psychosocial difficulties. The link between stigma and psychosocial difficulties is important because children with physical health conditions are already at a greater risk of mental health problems (Ferro et al., 2015; Hysing et al., 2007). However, it should be noted that the mean score for psychosocial difficulties in this study are relatively low (6.3 out of a possible 20). Although the standard deviation of 4 suggests that there was notable variability in the scores, a low mean level of psychosocial difficulties in this sample does mean some caution is required in interpreting these findings. Psychosocial difficulties were associated with both sub-scales of self-stigma and perceived stigma, as derived from PCA of the Child Stigma Scale. Emotional and social difficulties could be accounted for by perceived stigma due to concerns about peer rejection, isolation and bullying (Elliott et al., 2005; McMurray et al., 2001; Strauss et al., 2007), and by self-stigma based on distress caused by feeling different, abnormal or defective (Elliott et al., 2005; Velsor-Friedrich et al., 2004). This highlights the importance of professionals being cognisant of stigma experiences, and provides an opportunity in clinical practice for prevention or for the provision of appropriate support for psychosocial difficulties.

Alongside child stigma, parent stigma also emerged as an independent predictor of children's psychosocial difficulties. Since child and parent stigma were not associated with each other, parent stigma may relate to children's psychosocial difficulties via a different mechanism. One explanation is that parental stigma could affect the way parents interact with their children, through either conscious or unconscious mechanisms. Higher levels of parent stigma have previously been associated with lower levels of emotional support, a lower level of participation in family leisure activities, and behavioural problems in children with epilepsy (Austin et al., 2008; Carlton-Ford et al., 1997). There may also be other parental factors which were not investigated in this study, which are relevant to understanding the psychology of living with a health condition. These findings highlight the complexity of how stigma may

relate to psychosocial difficulties, and emphasises the importance of professionals listening to the accounts of both child and parental perceptions of stigma, even if these are seemingly discrepant with each other.

Negative illness perceptions were associated with higher levels of children's psychosocial difficulties, although it did not emerge as an independent predictor. It is possible that illness perceptions as a construct or the way it was measured in this study mean that it is not a useful construct in understanding children's psychosocial experiences. However, whilst the association between illness perceptions and psychosocial difficulties supports previous findings (Austin et al., 2006; le Coq et al., 2000; Wagner et al., 2008), illness perceptions were highly correlated with child stigma, suggesting that these two factors may account for some shared variance in children's psychosocial difficulties. This highlights the importance of considering these related factors together, rather than in isolation. This relationship between child stigma and illness perceptions could have useful implications in the context of psychosocial difficulties: illness perceptions fit into existing psychological health models such as the common sense model of illness representations (Diefenbach et al., 1996), and therefore provide a possible target for interventions.

Child concealment did not significantly predict children's psychosocial difficulties in this study. Despite previous studies which documented the negative consequences of concealment (Pachankis, 2007), these findings highlight how the 'disclosure decision' can be a challenging one. The qualitative literature has reported how children's decision to conceal their medical condition can be protective, due to the reality of stigmatising responses from others (Benson et al., 2015a; Christian et al., 1997; Elliott et al., 2005). The approach to concealment also varied depending on the target: for example by sharing with close friends, but not other peers (Kaushansky et al., 2017; McEwan et al., 2004; McMurray et al., 2001). However, the Child Secrecy Scale did not allow for measurement of these nuances in the disclosure dilemma, by

being focused only on children's beliefs about concealment. Future research could investigate patterns of disclosure or concealment in children with physical health conditions, depending on who the disclosure target is.

5.4. Limitations

There are a number of limitations to this study. First, the sample size was relatively small, meaning that there was not enough statistical power to control for multiple confounding variables in all regression analyses. Second, although a range of health conditions was included in order to make findings more applicable to a wider range of children, these findings may not generalise to all physical health conditions. Replication of these findings in a wider range of conditions with a larger sample size is required. Third, the measures of stigma and illness perceptions were developed for children with epilepsy, and the measure of concealment for children with mental health problems. Although most measures had been used with a range of other health conditions, they may not have captured all the experiences relevant to the construct they were measuring in a population of children with physical health conditions. Furthermore, there were limitations in how some of the measures operationalised constructs. For example, the CATIS could be argued to be a categorical measure due to the differences in responses for each item, and also uses frequency as an indicator of beliefs being more 'positive' or 'negative'. There is also an implicit assumption within the measure about what is considered to be the correct way to perceive illness, although this may vary between individuals, professionals and wider cultures. Fourthly, the analyses used in this study were based on linear relationships. However, the relatively low or neutral scores on many of the measures mean that it could have been useful to explore curvilinear relationships between variables in addition to the planned analyses, to investigate relationships between variables at the higher or lower ends of each distribution.

5.5. Implications

The association of both child and parental stigma with children's psychosocial difficulties highlights the importance of stigma awareness in professionals, and indicates that professionals should be enquiring about stigma experiences in order to identify which children may be at greater risk of psychosocial difficulties. However, overall low psychosocial scores in this study suggest that an assumption should not be made that all children would require additional psychological support. The perspectives of both children and parents should be considered as valuable information, even if their accounts appear to be incongruent with one another. Although child and parental stigma predicted their respective concealment of the child's health condition, concealment did not predict children's psychosocial difficulties. Professionals should therefore not assume that discouraging concealment will improve a child's psychosocial experience, but instead support families to consider the costs and benefits of this decision. These implications should be considered in the context of the above limitations, where further replication in a larger sample across a wider range of health conditions is required.

5.6. Future Research

The relationship between stigma and concealment could be used to inform future interventions. For example, existing interventions based on stigma and disclosure decisions in adolescents with mental health problems (Mulfinger et al., 2018) could be adapted for children with health conditions and their parents. Future research around the differences in concealment based on the target (e.g. close friends, peers, teachers) could provide a more nuanced understanding of the disclosure decisions made by children and their parents. Illness perceptions have already been included in existing psychological models (e.g. Diefenbach et al., 1996), and further investigation of the nature of the relationships between child stigma, illness perceptions and psychosocial difficulties could therefore help to inform future

interventions. Finally, future research could investigate what the primary influences are on children's stigma experiences, given that no associations between any parent and child-rated factors were found in this study. There is some emerging evidence that school-based interventions have shown shifts in attitudes towards children with epilepsy (Murthy et al., 2016), and the role of public stigma in children with health conditions provides an avenue for future research.

5.7. Conclusions

This study has highlighted the importance of stigma experiences as reported by both children with physical health conditions and their parents. Child and parental stigma were associated with child and parental concealment respectively, and also to children's psychosocial difficulties. The sources influencing the children's perceptions of stigma may be primarily external to their family, with no association found between child and parental factors. A greater awareness by healthcare professionals of stigma is key, so that families can be adequately supported in dealing with stigma, managing decisions about concealment, and in managing psychosocial difficulties.

6. References

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Part 3: Critical Appraisal

1. Introduction

This critical appraisal is divided into three sections, which offer reflections on aspects of the research process. The first part focuses on the investigation of different medical conditions: the rationale and personal reflections for including several conditions rather than focussing on one condition are presented, followed by a discussion of whether the variables in this study differed between conditions. The second part documents the process of selecting the measures in the empirical studies, and an evaluation of the strengths and limitations of these. The third part focuses on the process of data collection, including reflections on the response rate and the sample characteristics.

2. Investigating Different Health Conditions

2.1. Rationale and Personal Reflections

This study sought to understand stigma and related factors across different physical health conditions. This is in contrast with most studies in the physical health stigma literature, which generally either focus on one condition, or most commonly only include children with epilepsy. Although children may have variable experiences based on the nature of their physical health condition, the predominant approach of studies only investigating one condition means that it is difficult to draw conclusions about the common stigma experiences of these children. My perspective on this is likely to be influenced by my own experiences and training within the field of psychology. Psychological models for understanding physical health conditions such as the common sense model of illness representations (Diefenbach et al., 1996) and the disability stress coping model (Wallander et al., 1995) provide a general framework for understanding physical health conditions, rather than being based on specific conditions. Furthermore, my experience of working with physical health difficulties prior to conducting this research was in mental health settings, where a general approach might be used and adaptations made based on the specific medical

condition. Therefore, rather than focussing first on the experiences of certain medical conditions, I was keen to understand children's more general experiences of stigma.

However, deciding to investigate several health conditions has the consequence of increasing the level of heterogeneity within a sample. While including a greater range of conditions means that the study findings are generalisable to a greater proportion of the paediatric population, it also increases the degree of extraneous variability and therefore introduces a greater proportion of error variance. Several other factors were taken into account during the decision-making process. Firstly, being part of a joint project meant that the sample needed to be appropriate for the aims of both studies. My colleague Jemma was investigating differences in stigma based on the visibility of children's health conditions, which introduced a need to include variety in how visible a child's health condition was likely to be. Although this was not in line with my own research questions, the benefits of a joint project meant that our capacity for data collection was greatly improved. Secondly, the medical teams at the paediatric hospital were consulted. They were keen to minimise high levels of medical variation, with the rationale of wanting to understand certain groups better. A compromise was reached based on these factors, with the eventual recruitment strategy involving data collection from two medical specialties: dermatology and urology.

2.2. Differences between Health Conditions in this Study

In this study, scores on each measure were compared between physical health conditions and medical specialties (dermatology or urology). Although the main function of this was to allow for these factors to be controlled for in subsequent analyses, it provides some insight into whether the experiences of stigma, concealment and illness perceptions in children and their parents differed based on the child's condition. For both child stigma and parent stigma, no differences were detected between groups based on the child's health condition or the medical speciality. Similarly, there were no significant differences between the mean scores

on the Child Stigma Scale and the Parent Stigma scale when compared to the scores from a previous study of children with epilepsy (Austin et al., 2004). Similarly, the level of psychosocial difficulties encountered by children did not appear to differ based on their health condition, although children with a comorbid condition or more frequent hospital attendance had higher levels of psychosocial difficulties. This highlights how it might not always be the health condition itself that is important in influencing children's experiences: rather the consequences of the health condition. However, it is possible that due to the sample size of this study, there were differences between health conditions for stigma and psychosocial difficulties which were not detected.

In contrast, child concealment, parental concealment and illness perceptions all showed significant group differences in scores based on the child's health condition. Parental concealment and illness perceptions scores also differed significantly based on whether children attended dermatology or urology clinics. Parents reported higher levels of concealment when their child had urology conditions, which included bladder or kidney problems, compared with dermatological conditions of eczema, psoriasis and epidermolysis bullosa. The reason for this is unclear, although a reluctance of parents to bring up their children's bladder-related issues to healthcare professionals due to shame has previously been found (Cederblad et al., 2014). In accordance with findings that child and parental concealment were not associated, a different pattern of concealment emerged for children: the highest concealment scores were in children with psoriasis. Adolescents with psoriasis have documented concerns about their appearance and concealment (Randa et al., 2018), but it is not clear why this score would be higher than other dermatological conditions. Finally, children's illness perceptions were more positive in urology than dermatology.

The sample size of this study means that it is difficult to make any generalisations or draw any conclusions based on differences between health conditions, and to do so was not the purpose of this study. However, this provides a rationale for future

research investigating a range of health conditions rather than focussing on one specific condition, because it provides further insight into whether stigma and related factors in children and parents vary between conditions. If there are no differences between health conditions, then this would have helpful implications in the supporting development of general stigma interventions. However, if differences between health conditions do occur, then this could provide useful insight into which experiences increase or decrease experiences of stigma, concealment and illness perceptions in children and their parents.

3. Selection and Evaluation of Measures

The focus on epilepsy in the stigma literature of children with physical health conditions meant that the selection of possible validated measures was limited. The available measures had either been designed to capture stigma experiences of children with epilepsy, or would require adapting from adult or mental health stigma measures. The items of the Child Stigma Scale were developed based on findings from literature reviews of the stigma experiences of children with epilepsy (Austin et al., 2004), meaning that it may not have captured the stigma experiences of children with other physical health conditions. However, the scale has been adapted and used with children who have other physical health conditions such as inflammatory bowel disease, disorders of sex development, and sickle cell disease (Gamwell et al., 2018; Rolston et al., 2015; Wakefield et al., 2017), as well as in child mental health populations (Kaushik et al., 2017; Moses, 2009), indicating that it was applicable across different conditions. Another positive feature of the Child Stigma Scale was that it had been developed together with the Parent Stigma Scale, and these had been used together in previous studies (Austin et al., 2004; Benson et al., 2016; Ryu et al., 2015). Similarly, although the Child Attitudes Towards Illness Scale was developed for children with epilepsy (Austin et al., 1993), it had been used and validated with a number of conditions such as asthma, arthritis and diabetes (Ramsey

et al., 2016). Therefore, the measures which had been developed for children with epilepsy appeared to be the most valid measures.

The process of identifying measures also required a clear understanding of the different types of stigma described in the literature. A possible concern about the use of the Child Stigma Scale was that the questions appeared to cover elements of both perceived stigma and self-stigma. For this reason, principal components analysis was used in order to establish whether the items in the scale emerged as more than one component. The findings of the principal component analysis did reveal the possibility of two constructs, aligning to those of self-stigma and perceived stigma. Given that using the components separately in correlational analyses did not reveal any difference in which relationships were significant, the entire Child Stigma Scale was used in the analysis of the empirical study. However, it was interesting to note that the strength of the effect sizes did differ: perceived stigma in children had a stronger positive correlation with concealment, whereas self-stigma had a stronger negative correlation with children's illness perceptions. The development of a validated measure which differentiates between self-stigma and perceived stigma could be a useful avenue for future research, so that the relationships of these constructs with different factors can be further investigated.

Finding a measure of concealment also proved to be difficult due to the complexity of this concept. It had previously emerged in a number of qualitative studies (McEwan et al., 2004; Moffat et al., 2009; Walker et al., 2014), or had been measured using one item (e.g. Quinn et al., 2014). A review of disclosure practices in children with epilepsy and their parents had concluded that there was no appropriate measure being used in the literature at that time (Benson et al., 2015b). However, in the paediatric mental health stigma literature, a Secrecy Scale had been developed for children and adolescents (Kaushik et al., 2017; Moses, 2009) which was based on a validated measure in adults (Link et al., 1991; Link et al., 1997). The Secrecy Scale was

therefore adapted for this study, so that concealment could be measured in both children and their parents, and both displayed good internal consistency.

The findings from this empirical study revealed how stigma can predict concealment in both children and their parents, but also how concealment does not necessarily equate to greater psychosocial difficulties in children. This could be due to the complexity of the decision of whether to disclose or conceal a child's health condition (Benson et al., 2015b). However, the Secrecy Scale did not specify the target of the decision to conceal or disclose. In the qualitative literature, several studies have described children choosing a strategy of selective disclosure, for example by disclosing their condition to close friends but hiding it from other peers, whereas others concealed their condition from all of their friends (Benson et al., 2015a; McEwan et al., 2004; Wang et al., 2010). There were also dilemmas posed about how much to share with teachers at school, due to concerns about being treated differently (Christian et al., 1997; Moola et al., 2011). Therefore, future research could build on the findings of this study by investigating whether these findings are consistent depending on the target of disclosure/ concealment. Given the lack of association between child and parental concealment in this study, comparing child and parental attitudes to concealment depending on the target of this decision could also help to indicate where these discrepancies may lie. For example, whether there is a difference between parents and children in their views about disclosing to peers and to teachers. This could provide useful insight into help-seeking behaviour, and identify areas where children may not be accessing support.

The issues encountered in this study when choosing appropriate measures to investigate stigma experiences in children with physical health conditions may also be representative of a barrier faced by researchers when pursuing this area of research. While the studies of children with epilepsy provide a good basis for future research in children with other physical health conditions, it means that the tools

available are limited. It is important that stigma research in the paediatric population is applied to a broader range of health conditions, so that there is not an inequity of experience caused by the nature of a child's health condition.

4. Data Collection

4.1. Response Rates

The overall response rate for this study was 25%, which was lower than expected. One of the major challenges encountered as part of this process was getting in touch with families: it is notable that of those who did not participate, 38% could not be contacted by telephone after an initial letter had been sent by post. Given that these families may have been interested in taking part, it is useful to consider what the barriers might have been to contacting these families. The vast majority of calls to families were made during traditional working hours, between 9am – 5pm. It is likely that for some parents, taking calls during the day might have been difficult as a result of work, caring, or childcare responsibilities. Making better use of technology could have been a useful way of overcoming this barrier, for example by contacting parents by email instead, which would have provided families with more agency about when the communication took place. Alternatively, making more calls to family outside of traditional working hours might have increased the likelihood of people picking up their phones.

There were also a substantial number of families who declined participation, forming 26% of the parents who were approached. Although no one reported stigma as a reason for not taking part, it is possible that stigma influenced parent's decisions about participation. One of the findings of the empirical study was that higher levels of parental stigma were associated with great concealment by parents of their child's health condition. In the same way that parents can act as a 'gateway' to services for their children (Chavira et al., 2017), it is possible that parents with higher levels of stigma may have influenced their decision about whether their child would take part

in this research project. Some parents declined participation on the basis that the study did not appear to be relevant to their child. However, the results from the empirical paper suggest that parents and children may have different perceptions about the level of stigma experienced by children. Although the lack of participation from these families means that it was not possible to know whether this was the case, the findings from this study can be used to understand possible barriers to participation in research as well as barriers to accessing support from services.

When the transition was made to remote data collection as a result of Covid-19, different challenges were encountered. It has been well documented that response rates can be lower when questionnaires are sent to participants to be completed remotely (e.g. Benfield et al., 2006; Ebert et al., 2018). This phenomenon was also found in this study, where 18 of the 30 parents who agreed to take part remotely did not return the questionnaires. Furthermore, the format of the questionnaire had been designed for face to face data collection, meaning that it was not possible to benefit from some of the advantages of remote data collection such as automated data entry. Transitioning from face-to-face to remote data collection also meant that heterogeneity in the form of data collection method was introduced, although analyses revealed no difference in any of the measures as rated by children or their parents between those who had completed the questionnaires remotely or face-to-face.

4.2. Sample Characteristics

There were a number of strengths and weaknesses of how representative the sample was in the empirical study. The number of children who identified as female and male was nearly equal (31 and 30 respectively), which was a strength of the study due to being representative of the general population. However, the large majority of parents were female. One of the unexpected findings was that children's psychosocial difficulties were rated as higher by parents who identified as female as opposed to those who identified as male. Discrepancies between the ratings of mothers and

fathers on psychiatric outcomes have previously been reported (Kazdin et al., 1983), but might have been less apparent if there had been a larger sample of fathers. Finally, in this study, 75.4% of children identified as White British/ White Other, 11.5% identified as Asian/ Asian British, 6.6% identified as Black/ Black British, and 6.6% as 'other' or preferred not to provide their ethnicity. The paediatric hospital where this study took place was based on London, but had a national remit. However, the ethnicity of children in this study is not representative of either local or national figures. According to the 2011 census (ONS, 2011), 60% of people in London identified as White British/ White Other, 20% identified as Asian/ Asian British, and 13% identified as Black British. In England and Wales, the 2011 census data showed that 86% of the population identified as White, 7.5% identified as Asian/ Asian British, and 3.3% identified as Black/ Black British. Although the ethnicity of children is more closely aligned with national figures, children who identified as Black, Asian or other ethnic backgrounds were still underrepresented in this study. Furthermore, health inequalities between ethnic categories are well documented, with greater prevalence of health conditions in Black and Asian people (Evandrou et al., 2016; Randhawa, 2007). This means that it is particularly important to strive for samples to be representative. Although no differences were detected on any of the measures based on the child's ethnic group, it is possible that this was due to the small number of children identifying as Black, Asian or other minority groups.

5. Conclusions

This critical appraisal sought to reflect on issues and learning points which arose over the course of conducting this research. Although greater heterogeneity was introduced as a result of deciding to look at a range of medical conditions, this allowed for an examination of whether there were differences between medical conditions for child and parent factors in this study. A previous focus in the literature on epilepsy led to some limitations of the available measures, which could present a barrier for future

research. Finally, a reflection on data collection considered some of the issues which may have contributed to a low response rate, and highlighted areas in which this sample could have been more representative. These reflections provide useful learning points for future research in this area.

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Appendix A: Full Parent Survey

BACKGROUND INFORMATION

PARTICIPANT ID: _____

1) Child's age:

Age: yearsmonths

2) Please state your child's gender identity:

.....

3) School year:

- | | | |
|---------------------------------|----------------------------------|----------------------------------|
| <input type="checkbox"/> Year 3 | <input type="checkbox"/> Year 4 | <input type="checkbox"/> Year 5 |
| <input type="checkbox"/> Year 6 | <input type="checkbox"/> Year 7 | <input type="checkbox"/> Year 8 |
| <input type="checkbox"/> Year 9 | <input type="checkbox"/> Year 10 | <input type="checkbox"/> Year 11 |

4) How would you describe your child's ethnicity? Please tick ONE box

A White

- British
 Any Other White background, *please write in*

B Mixed

- Any Mixed background, *please write in*

C Asian/Asian British

- Indian
 Pakistani
 Bangladeshi
 Chinese
 Any Other Asian background, *please write in*

D Black, Black British

- Caribbean
 African
 Any Other Black background, *please write in*

E Other ethnic group

- Any Other background, *please write in*

F Prefer not to say

5) Please state your age:

Age: years

6) Please state your gender identity:

.....

7) How would you describe your ethnicity?

Please tick ONE box

A White

- British
 Any Other White background, *please write in*

B Mixed

- Any Mixed background, *please write in*

C Asian/Asian British

- Indian
 Pakistani
 Bangladeshi
 Chinese
 Any Other Asian background, *please write in*

D Black, Black British

- Caribbean
 African
 Any Other Black background, *please write in*

E Other ethnic group

- Any Other background, *please write in*

F Prefer not to say

MEDICAL CONDITION

1) What is the name of the condition for which your child comes to GOSH?

.....

2) Does your child have any other medical or developmental problems?

Yes

No

If yes, please let the researcher know before proceeding further:

.....

3) Is your child currently, acutely unwell and/or requiring immediate medical attention?

Yes

No

If yes, please let the researcher know before proceeding further:

.....

5) How old was your child when the medical condition began?

Present at birth

Less than 5 years

5 years or older

6) On average, how often does your child attend hospital in a year?

1-2 times a year

3-10 times

More than 10 times

7) Can your child's medical condition be seen by someone who walks past them on the street?

Yes

No

Child Psychological Well-being

For each item, please mark the box for Not True, Somewhat True or Certainly True. It would help us if you answered all items as best you can even if you are not absolutely certain or the item seems daft! Please give your answers on the basis of the child's behaviour over the last six months.

	Not True	Somewhat True	Certainly True
1. Considerate of other people's feelings			
2. Restless, overactive, cannot stay still for long			
3. Often complains of headaches, stomach-aches or sickness			
4. Shares readily with other children (treats, toys, pencils, etc.)			
5. Often has temper tantrums or hot tempers			
6. Rather solitary, tends to play alone			
7. Generally obedient, usually does what adults request			
8. Many worries, often seems worried			
9. Helpful if someone is hurt, upset or feeling ill			
10. Constantly fidgeting or squirming			
11. Has at least one good friend			
12. Often fights with other children or bullies them			
13. Often unhappy, down-hearted or tearful			
14. Generally liked by other children			
15. Easily distracted, concentration wanders			
16. Nervous or clingy in new situations, easily loses confidence			
17. Kind to younger children			
18. Often lies or cheats			
19. Picked on or bullied by other children			
20. Often volunteers to help others (parents, teachers, other children)			
21. Thinks things out before acting			
22. Steals from home, school or elsewhere			
23. Gets on better with adults than with other children			
24. Many fears, easily scared			
25. Sees tasks through to the end, good attention span			

How other people view my child's medical condition

	Strongly disagree	Disagree	Neither	Agree	Strongly agree
1. People who know that my child has a medical condition treat him/her differently					
2. It doesn't really matter what I say to people about my child's medical condition, they usually have made their minds up					
3. My child always has to prove him/herself because of their medical condition					
4. Because of the medical condition, my child will have problems in finding a husband or wife					
6. In many people's minds, a medical condition attaches a stigma or a label to my child.					

Helping my child to cope with how other people view their medical condition

	disagree a lot	disagree	agree	agree a lot
1. There is no reason for a child to hide the fact that he or she could be receiving help for a medical condition				
2. I usually wait until I know a person really well before I tell them if my child is receiving help for a medical condition				
3. When I meet people for the first time, I make a special effort to keep the fact that my child is receiving help for their medical condition to myself				
4. I often worry that someone will tell others about my child's medical condition without my permission				
5. I feel like I need to hide the fact that my child has a medical condition from other children their age				
6. I often feel the need to hide the fact that my child is receiving help for their medical condition.				
7. If a child is getting help with their medical condition, the best thing to do is keep it to yourself.				

END OF QUESTIONS

PLEASE RETURN FORMS TO RESEARCHER

THANK YOU VERY MUCH FOR YOUR PARTICIPATION!

Appendix B: Full Child Survey

PARTICIPANT ID:

**What is the medical or surgical condition that you come to GOSH for?
e.g., heart condition? Eczema? Port-wine stain? bladder problems?**

Please write it here:.....

Please answer these questions about having *[insert named condition here]*.

1. How good or bad do you feel it is that you have this condition?

Very good A little good Not Sure A little bad Very bad

2. How fair is it that you have a medical condition?

Very fair A little fair Not sure A little unfair Very unfair

3. How happy or sad is it for you to have a medical condition?

Very sad A little sad Not sure A little happy Very happy

4. How bad or good do you feel it is to have a medical condition?

Very good A little good Not Sure A little bad Very bad

5. How often do you feel that your medical condition is your fault?

Never Not often Sometimes Often Very often

6. How often do you feel that your medical condition keeps you from doing things you like?

Very often Often Sometimes Not often Never

7. How often do you feel that you will always be sick?

Never Not often Sometimes Often Very often

8. How often do you feel that your medical condition keeps you from starting new things?

Very often Often Sometimes Not often Never

9. How often do you feel different from others because of your medical condition?

Never Not often Sometimes Often Very often

10. How often do you feel bad because you have a medical condition?

Very often Often Sometimes Not often Never

11. How often do you feel sad about being sick?

Never Not often Sometimes Often Very often

12. How often do you feel happy even though you have a medical condition?

Never Not often Sometimes Often Very often

13. How often do you feel just as good as other kids your age even though you have a medical condition?

Very often Often Sometimes Not often Never

How do other people view your *[insert named condition here]*?

	Never	Not Often	Sometimes	Often	Very Often
1. How often do you feel different from other kids because you have a medical condition?					
2. How often do you feel people may not like you if they know you have a medical condition?					
3. How often do you feel other children are uncomfortable with you because of your medical condition?					
4. How often do you feel people may not want to be friends with you if they know you have a medical condition?					
5. How often do you feel people would not want to go out with you or ask you to parties if they know you have a medical condition?					
6. How often do you feel embarrassed about your medical condition?					
7. How often do you keep your medical condition a secret from other kids?					
8. How often do you try to avoid talking to other people about your medical condition?					

How I cope with other people's reactions to my *[insert name of condition here]*

	disagree a lot	disagree	agree	agree a lot
1. There is no reason for a person to hide the fact that he or she could be receiving help for a medical condition				
2. I usually wait until I know a person really well before I tell them if I am receiving help for a medical condition				
3. When I meet people for the first time, I make a special effort to keep the fact that I am receiving help for my medical condition to myself				
4. I often worry that someone will tell others about my medical condition without my permission				
5. I feel like I need to hide the fact that I have a medical condition from children my age				
6. I often feel the need to hide the fact that I am receiving help for my medical condition.				
7. If you are getting help with your medical condition, the best thing to do is keep it to yourself.				

Appendix C: Study Invitation Letter & Participant Information Sheets

Great Ormond Street
Hospital for Children



Great Ormond Street
London WC1N 3JH

NHS Foundation Trust

Tel: 020 7405 9200

“Date”

“Name and Address of Recipient”

To the Parent/Guardian of *name of patient*

Re: Stigma and Psychological Well-being in Children and Young People with Chronic Medical Conditions (V.1)

We would like to invite you and your child to take part in a questionnaire study on stigma in children and young people (CYP) with medical conditions. Stigma is the feeling of being different to everyone else in a negative way. We would like to find out whether GOSH patients feel that having a medical condition causes them to feel different in this way. We would also like to understand if feeling different because of a medical condition might affect how the young person lives with their medical condition and how it affects their psychological well-being.

As psychologists at GOSH, we are keen to learn more about how young people feel about having a medical condition. We hope that this research will help us to help our patients feel less bothered about having a medical condition; to feel more confident about revealing or talking about their condition with their family, friends and people they come across in their daily lives; to develop positive help-seeking behaviours so that they receive the best healthcare and to feel more confident about themselves generally.

There are two information sheets attached to this letter that explain our project in more detail. One is for parents/guardians and the other is for the young person. The information sheets explain what you and your child would be doing if you agree to take part in this project.

Please read the information sheets carefully. A member of the GOSH team will be in touch with you by telephone in 1-2 weeks' time to answer any further questions you may have and to find out if you and your child are interested in taking part.

You are very welcome to contact us on the telephone number below if there's anything that we can do to help you to understand this project better.

Yours sincerely,



Dr. Kristina Soon

Clinical Psychologist for Dermatology/Lead Investigator



Appendix D: Participant Information Sheet (Children)

Participant Information Sheet: Patients 8-14 years old

Study Title: How does having a medical condition affect how children and young people feel about themselves?

We would like to see if you and your parents/guardians would like to take part in our research study.

Before you decide if you would like to join in, we would like you to understand what our study is about and what you have to do if you take part.

Please read the information below. If anything is unclear, please feel free to discuss it with your parents/guardians. We have sent them some information about our study too.

One of us will be phoning you and your parents/guardians in a few days' time. We can answer any questions or worries you have about taking part then.

Our telephone number and email address are written at the bottom of this information sheet. You can phone us with your questions if you prefer.

Why are we doing this research?

We know that having a serious medical condition can be difficult for children and young people. Some young people who have medical conditions have told us that they worry about what other people might think about them, if they found out that they have a medical condition. This can cause them to worry, to hide their medical condition or to feel shy around other people. We want to help patients at GOSH feel more confident about talking about their medical condition, to not hold in their worries and to feel happier in themselves.

Why have you been invited to take part?

You and your parents/guardians have been invited to take part because you are a patient at GOSH and we would like to find out more about what it is like for you to have a medical condition.

Do I have to take part?

You do not have to take part in this study. It is up to you!

No-one will be upset or angry. Your doctors and nurses will still work with you in the same way and do the best that they can to keep you well.

What will happen if I take part?

If you agree to take part, a time will be arranged with your parents (probably the next time you come to GOSH for an appointment) for one of us to meet with you. When we meet with you we will check that you are still happy to take part (You can say no if you've changed your mind). If you are happy to continue, we will give you some questionnaires to complete on an electronic tablet. Your parents/guardians and the researcher will be there to help you if you have any questions about what to do. The questionnaire will take you about 10-20 minutes to complete.

Could anything bad happen if I take part?

You only need to fill out a questionnaire with your parents/guardians nearby. As such, we don't think it is likely that anything bad can happen. Sometimes when people fill out questionnaires about personal things, it can be a bit sad. So it is possible that you may feel a bit sad when you fill out the questionnaires. If you do, your parents/guardians and the researcher are there to support you and to help you to feel better if necessary.

Will taking part in this study help me?

Taking part might not help you. However, some people who fill out our questionnaires tell us that it can help them to understand their own feelings a bit better and help them to talk to others about their feelings. We also hope that this study will help us to improve how we look after patients at GOSH so you might be helped by these improvements in the future.

How will taking part in this study help others?

Helping us to understand how it feels to have a medical condition will help us to look after the feelings of all patients at GOSH who might feel the same way. We hope that this study will improve how we look after patients in GOSH so that they can feel really good about themselves.

Who can I ask if I have questions about this study.

If you have questions or worries about this study you can:

- Ask your parents/guardians. They have also been given information about this study and they might be able to help.
- You or your parents/guardians can contact the lead researcher on this study:
Dr Kristina Soon, [REDACTED]
[REDACTED]

If you have any concerns about the conduct of this study you can contact GOSH Patient Advice and Liaison Service (Pals) on 020 7829 7862 or pals@gosh.nhs.uk

Appendix E: Participant Information Sheet (Parents)

Participant Information Sheet: Parents/Guardians

Study Title: How does having a medical condition affect how children and young people feel about themselves?

We would like to invite you and your child to take part in our research study.

Before you decide if you would like to join in, we would like you to understand what our study is about and what you and your child have to do if you take part.

Please read the information below. One of us will try to contact you by telephone in a few days' time. We can answer any questions or worries you have about taking part then.

Our telephone number and contact email address are written at the bottom of this information sheet. You can phone or email us with your questions if you prefer.

Feel free to discuss this study with your friends and family or healthcare professionals.

Why are we doing this research?

We know that having a serious medical condition can be difficult for children and young people. Some young people who have medical conditions have told us that they worry about what other people might think about them, if they found out that they have a medical condition. This can cause them to worry, to hide their medical condition or to feel shy around other people. It can also result in the patient not wanting to seek medical care for their condition because they feel uncomfortable about talking about it or drawing attention to it. Avoiding medical care can have a negative impact on the patient's health and wellbeing.

We want to help patients at GOSH feel more confident about talking about their medical condition, to not hold in their worries, to feel happier in themselves and to be able to work proactively with healthcare professionals to get the best treatment outcome. By collecting this information, we hope to be able to understand the emotional needs of our patients better and to be able to provide care that is supportive to those emotional needs as well as their medical needs.

Who is eligible to take part?

We are approaching young people aged eight to 14 years, who have dermatological or urological conditions, who are cared for at Great Ormond Street Hospital. We would also like at least one of their parents/guardians to take part.

Do we have to take part?

No. It is entirely up to you and your child to decide if you want to participate. You can also change your mind at any point if you don't want to continue. Your decision will have no bearing on your child's ongoing clinical care at GOSH. We will continue to do our best for you and your child.

What are we looking at in particular?

We want to find out about how the young person feels about having their medical condition, what they think other people feel about them having a medical condition and whether or not they try to hide their condition from others. We will be asking parents very similar questions about how they feel about their child having a medical condition, how others feel about your child having a medical condition and how they manage that. Also, we will look at whether how obvious the medical condition is to other people has an impact on how the young person feels.

What would you and your child have to do?

Parents and children will be asked to complete a few questionnaires on an electronic tablet the next time you come to GOSH for an appointment. A member of the research team will arrange to meet you and will help you with this if necessary. We estimate that an eight-year-old of average reading ability will take no more than 20 minutes to complete the questionnaire. We estimate that the parent will take no more than 10-15 minutes to complete their questionnaires.

Where would this happen?

In order to minimise inconvenience to you and your child, we would like to meet you at GOSH on a day when you are attending an out-patient appointment here. A member of the research team can arrange a time to meet with you that is convenient so as not to interfere with you attending the appointment. The researcher will have access to a private area in the hospital, such as a clinic room, for you and your child to fill in the questionnaires.

How long will the study run for?

Each participant will only need to complete one set of questionnaires one time. Therefore, for each participant, their involvement will be over within 20-30 minutes. The study itself will run for approximately six months or until about 65 young patients and their parents have taken part. We aim to contact patients and their families between August 2019 and March 2020.

Are there any risks involved in taking part?

Because taking part in this study only involves completing a questionnaire, we don't think that it is likely that anything bad will happen. Sometimes, when people fill out questionnaires about personal thoughts, feelings and experiences, they can start to feel quite emotional. You and your child are welcome to stop if you feel too upset. The researcher, who is a qualified psychologist, is there to support you and your child if this happens.

Are there any benefits in taking part?

The main aim of this study is to help to develop our clinical services at GOSH. As such, we do not anticipate that you and your child will benefit directly from taking part. However, participants in our previous studies have told us that filling out this type of questionnaire can help them to understand their own feelings a bit better and to feel ok about having those feelings. After taking part in a study like this, the patient or parent might feel that meeting with a trained mental health practitioner would be helpful. The researcher can discuss options for further psychological input with you.

How will taking part in this study help others?

Helping us to understand how it feels to have a medical condition will help us to develop our services so that we can help all patients at GOSH to feel better about who they are and about their medical condition.

How will the information that we share with you be protected?

Participant confidentiality is very important to us. As such, all information that we gather will be stored using a code number for each participant instead of their name so that it cannot be linked to individual patients or parents. The information will be stored electronically on the GOSH network which has very high standards of security, for 15 years, in line with the EU General Data Protection Regulations (GDPR) and the Data Protection Act (2008).

A note will be made in your child's GOSH patient records that they have participated in this study. The details of their involvement and their questionnaire responses will not be stored in their patient record.

If you withdraw from the study, we will keep and continue to use all the data that we have already collected from you and your child. We will not collect any further data.

What should I do if I wish to make a complaint?

If you have concerns about any aspect of this study you should speak to the lead investigator of the research team in the first place:

Dr Kristina Soon

Clinical Psychologist, Lead Investigator

[REDACTED]

[REDACTED]

If your concerns are still unresolved, you can contact:

GOSH Patient Advice and Liaison Service (Pals)

020 7829 7862

pals@gosh.nhs.uk

What will happen to the results of this study?

When the study is completed we will share our findings with GOSH healthcare professionals. We will present our study at a conference for healthcare professionals and we will publish the study in a professional journal. All results shared will be anonymous and will not identify individual participants.

You will also be asked, at the end of your participation if you would like to receive a summary of the study findings at the conclusion of the project. If you would, you can provide your preferred contact details (either email address or mailing address) to the researcher who will send you the report in due course.

Who is organising and funding this study?

This study is a collaboration between researchers from Great Ormond Street Hospital and University College London. The researchers will not be receiving any extra money, over and above their normal salary, for conducting this research.

Who has reviewed this study to make sure that it is of sufficient quality?

This study has gone through several reviews. It has been approved by the NHS Health Research Association (Registration No:), and the Great Ormond Street Hospital Clinical Research Adoptions Committee (Registration No...).

Expenses and Payments?

We do not anticipate that patients and families will incur any extra costs in participating in this study over and above the usual costs of attending an appointment at GOSH. As such, we will not be providing any payments for participation.

What do I do now?

Talk to your child to discuss whether you both wish to take part in this study. You can also see what else you and/or your child would like to know about the study before you decide whether to take part.

One of the research team will be phoning you within a week of you receiving this information sheet. They can answer your questions. You can let them know if you would like to take part or not.

Thank you for taking the time to read this information sheet

HRA Information Governance Transparency Statement

Great Ormond Street Hospital for Children NHS Trust (GOSH) is the sponsor for this study based in UK. We will be using information from you and your child in order to undertake this study and will act as the data controller for this study. This means that we are responsible for looking after your information and using it properly. GOSH will keep identifiable information about you and your child until 6-12 months after the end of this study after which all identifiable information will be deleted.

Your rights to access, change or move your information are limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, we will keep the information about you that we have already obtained. To safeguard your rights, we will use the minimum personally-identifiable information possible.

GOSH will keep your child's name, hospital number and contact details confidential and will not pass this information to anyone else. GOSH will use this information as needed, to contact you about the research study, and make sure that relevant information about the study is recorded for your care, and to oversee the quality of the study. Certain individuals from GOSH and regulatory organisations may look at your child's medical and research records to check the accuracy of the research study. The people who analyse the information will not be able to identify you or your child and will not be able to find out your or your child's name, hospital number or contact details.

You can find out more about how we use your information by contacting

Appendix F: Assent Form for Children

Great Ormond Street Hospital for Children



NHS Foundation Trust

Participant Identification Number:

ASSENT FORM (Child/Young Person Participation)

Great Ormond Street
London WC1N 3JH

Tel: 020 7405 9200

**Title of Project: How does having a medical condition affect
how children and young people feel about themselves?**

Name of Researcher:(Please print clearly)

Please
initial
box:

1. I confirm that I have read the information sheet dated..... (version.....) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.
2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my medical care or legal rights being affected.
3. I understand that data collected during the study, may be looked at by researchers employed by the NHS where it is relevant to my taking part in this research. I give permission for these individuals to have access to my data.
4. I understand that the information collected about me will be stored anonymously
5. I agree that my participation in the study will be noted in my GOSH medical records.
6. I agree to take part in the above study.

Name of Participant

Signature

Date

Appendix G: Consent Form for Parent on Behalf of Child

Great Ormond Street Hospital for Children

NHS Foundation Trust

Participant Identification Number:

CONSENT FORM

(Parent/Guardian on behalf of Child)

Great Ormond Street
London WC1N 3JH

Tel: 020 7405 9200

Title of Project: How does having a medical condition affect how children and young people feel about themselves?

Name of Researcher:(Please print clearly)

Please
initial box:

1. I confirm that I have read the information sheet dated.....
(version.....) for the above study. I have had the opportunity to consider
the information, ask questions and have had these answered satisfactorily.
2. I understand that my child's participation is voluntary and that they are free
to withdraw at any time without giving any reason, without my child's medical
care or legal rights being affected.
3. I understand that data collected during the study, may be looked at by
researchers employed by the NHS where it is relevant to my taking part in
this research. I give permission for these individuals to have access to my
data.
4. I understand that the information collected about my child will be stored
anonymously
5. I agree that my child's participation in the study will be noted in their GOSH
medical records
6. I agree to take part in the above study.
7. I wish to be sent information about the results of the study

My preferred contact address is.....

Name of Participant

Name of Signatory

Relationship to Participant

Date

Signature

Appendix H: Consent form for Parents

Great Ormond Street Hospital for Children



NHS Foundation Trust

Participant Identification Number:

London WC1N 3JH

Tel: 020 7405 9200

CONSENT FORM

(Parent/Guardian Participation)

Title of Project: How does having a medical condition affect how children and young people feel about themselves?

Name of Researcher:(Please print clearly)

Please
initial box:

1. I confirm that I have read the information sheet dated.....
(version.....) for the above study. I have had the opportunity to consider
the information, ask questions and have had these answered satisfactorily.
2. I understand that my participation is voluntary and that I am free to withdraw
at any time without giving any reason, without my child's medical care or
legal rights being affected.
3. I understand that data collected during the study, may be looked at by
researchers employed by the NHS where it is relevant to my taking part in
this research. I give permission for these individuals to have access to my
data.
4. I understand that the information collected about me will be stored
anonymously.
5. I agree that my child's participation in the study will be noted in their GOSH
medical records
6. I agree to take part in the above study.

Name of Participant

Name of Signatory

Relationship to Participant

Date

Signature

Appendix I: Ethical Approval Letters



**Health Research
Authority**

London - Central Research Ethics Committee

3rd Floor, Barlow House
Minshull Street
Manchester
M1 3DZ

Telephone: 0207 1048 007

Please note: This is the favourable opinion of the REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval

29 August 2019

Dr Kristina Soon, Highly Specialist Clinical Psychologist

[REDACTED]
[REDACTED]
[REDACTED]
[REDACTED]

Dear Dr Soon

Study title:	Stigma, concealment and psychological wellbeing in children and young people with chronic medical conditions.
REC reference:	19/LO/0967
Protocol number:	N/A
IRAS project ID:	256531

Thank you for your letter of 19 August 2019, responding to the Committee's request for further information on the above research and submitting revised documentation. The further information has been considered on behalf of the Committee by the Chair.

Confirmation of ethical opinion

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

Conditions of the favourable opinion

The 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

It is a condition of the REC favourable opinion that all clinical trials are registered on a publicly accessible database. For this purpose, clinical trials are defined as the first four project categories in IRAS project filter question 2. For clinical trials of investigational medicinal products (CTIMPs), other than adult phase I trials, registration is a legal requirement.

Registration should take place as early as possible and within six weeks of recruiting the first research participant at the latest. Failure to register is a breach of these approval conditions, unless a deferral has been agreed by or on behalf of the Research Ethics Committee (see here for more information on requesting a deferral: <https://www.hra.nhs.uk/planning-and-improvingresearch/research-planning/research-registration-research-project-identifiers/>)

As set out in the UK Policy Framework, research sponsors are responsible for making information about research publicly available before it starts e.g. by registering the research project on a publicly accessible register. Further guidance on registration is available at: <https://www.hra.nhs.uk/planning-and-improving-research/research-planning/transparencyresponsibilities/>

You should notify the REC of the registration details. We will audit these as part of the annual progress reporting process.

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

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

Ethical review of research sites

NHS/HSC sites

The favourable opinion applies to all NHS/HSC sites listed in the application 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

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:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Covering letter on headed paper [Cover Letter]	1	13 May 2019
IRAS Application Form [IRAS_Form_14052019]		14 May 2019
IRAS Application Form XML file [IRAS_Form_14052019]		14 May 2019
IRAS Checklist XML [Checklist_14052019]		14 May 2019
IRAS Checklist XML [Checklist_29072019]		29 July 2019
IRAS Checklist XML [Checklist_19082019]		19 August 2019
Letters of invitation to participant [Letter of invitation]	1	13 May 2019
Other [Assent Form CYP 8-14yrs]	2	13 May 2019
Other [CRAC outcome letter March 209]	1	11 March 2019
Other [Questionnaires Parent: Guardian V4]	4	19 August 2019
Other [Applicant Response to Provisional Opinion]	2	19 August 2019
Participant consent form [Parent Consent for child V 3]	3	19 August 2019
Participant consent form [Parent Consent for Self V4]	4	19 August 2019
Participant information sheet (PIS) [Participant Information Sheet 8 - 14 V4]	4	19 August 2019

Participant information sheet (PIS) [Participant Information Sheet Parents/Guardians V5]		19 August 2019
Referee's report or other scientific critique report [CRAC outcome letter May 2019]	1	10 May 2019
Research protocol or project proposal [Study protocol]	2	13 May 2019
Summary CV for Chief Investigator (CI) [CI resume]	1	13 May 2019
Summary, synopsis or diagram (flowchart) of protocol in non-technical language [Data Collection Protocol V2]	2	13 May 2019
Validated questionnaire [Questionnaires CYP]	1	13 May 2019

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/qualityassurance/>

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-improvingresearch/learning/>

19/LO/0967

Please quote this number on all correspondence

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

Yours sincerely



pp

Dr Andrew Hilson Chair

Email: NRESCommittee.London-Central@nhs.net

Enclosure: "After ethical review – guidance for researchers"

Copy to: Ms Vanshree Patel

Dr Kristina Soon

Highly Specialist Clinical Psychologist

Email: hra.approval@nhs.net

HCRW.approvals@wales.nhs.uk



03 October 2019

Dear Dr Soon

HRA and Health and Care

Study title:	Stigma, concealment and psychological wellbeing in children and young people with chronic medical conditions.
IRAS project ID:	256531
Protocol number:	N/A
REC reference:	19/LO/0967
Sponsor	Great Ormond Street Hospital for Children NHS Trust

I am pleased to confirm that [HRA and Health and Care Research Wales \(HCRW\) Approval](#) has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

Please now work with participating NHS organisations to confirm capacity and capability, in line with the instructions provided in the “Information to support study set up” section towards the end of this letter.

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?

HRA and HCRW Approval does not apply to NHS/HSC organisations within Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance

report (including this letter) have been sent to the coordinating centre of each participating nation. The relevant national coordinating function/s will contact you as appropriate.

Please see [IRAS Help](#) for information on working with NHS/HSC organisations in Northern Ireland and Scotland.

How should I work with participating non-NHS organisations?

HRA and HCRW Approval does not apply to non-NHS organisations. You should work with your non-NHS organisations to [obtain local agreement](#) in accordance with their procedures.

What are my notification responsibilities during the study?

The standard conditions document "[After Ethical Review – guidance for sponsors and investigators](#)", issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including:

- Registration of research
- Notifying amendments
- Notifying the end of the study

The [HRA website](#) also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

Who should I contact for further information?

Please do not hesitate to contact me for assistance with this application. My contact details are below.

Your IRAS project ID is **256531**. Please quote this on all correspondence.

Yours sincerely,

Rekha Keshvara

Approvals Manager

Email: hra.approval@nhs.net

Copy to: *Ms Vanshree Patel* **List of Documents**

The final document set assessed and approved by HRA and HCRW Approval is listed below.

<i>Document</i>	<i>Version</i>	<i>Date</i>
Covering letter on headed paper [Cover Letter]	1	13 May 2019
IRAS Application Form [IRAS_Form_14052019]		14 May 2019
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Summary, synopsis or diagram (flowchart) of protocol in non technical language [Data Collection Protocol V2]	2	13 May 2019
Validated questionnaire [Questionnaires CYP]	1	13 May 2019

10/05/2019

PI: Kristina Soon

R&D number: 19SH02

Title: Stigma, concealment and psychological wellbeing in children and young people

(CYP) with chronic medical conditions

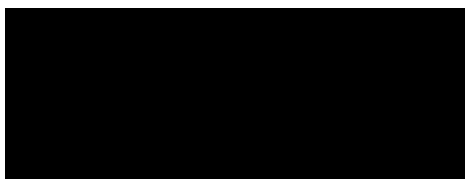
Dear Kristina,

Thank you for your response to the CRAC outcome letter dated 11/3/19. The Committee is satisfied that any concerns have now been addressed and has no objections to the conduct of this project at GOSH.

You will shortly be contacted by R&D Governance who will support you through the process of obtaining the necessary approvals before your project can begin. You must not commence your project before receiving R&D approval. Please find attached further information regarding the next stages in the research administration process.

Decision: Approval

Regards,



Dr Owen Arthurs

Chair

Clinical Research Adoption Committee

Joint Research and Development Office
Division of Research and Innovation
UCL Institute of Child Health, 30 Guilford Street, London WC1N 1EH
Tel: 020 7905 2700 Fax: 020 7905 2201
www.gosh.nhs.uk

The child first and always

Appendix J: Joint Thesis Contribution Statement

This was a joint thesis project, conducted together with Jemma Ambrose, who was investigating the stigma experiences of children with visible and less visible physical health conditions (Ambrose, 2020).

The systematic literature review documented in Part 1 was carried out independently. Jemma Ambrose acted as a second rater using the CASP framework for ten of the nineteen studies, as described in the methodology.

For the empirical study documented in Part 1, the recruitment strategy for the empirical study was jointly planned and the process of data collection was shared equally. All subsequent analyses were conducted independently.

Appendix K: Preliminary analyses of demographic and clinical factors

Demographic Variables and Test Variable – Independent samples t-tests

Child Stigma					
<i>Demographic Variable</i>	<i>n</i>	<i>M</i>	<i>SD</i>	<i>t</i>	<i>p</i>
Child Gender				1.52	.14
Female	31	2.57	.94		
Male	30	2.21	.92		
Medical Specialty				-2.00	.05
Dermatology	34	2.59	.92		
Urology	27	2.12	.91		
Other Medical Condition				.47	.64
Yes	18	2.48	.91		
No	43	2.36	.47		
Parent Gender				1.72	.24
Female	51	19.63	7.76		
Male	10	16.60	5.52		
Mode of data collection				1.01	.32
Face to face	49	2.45	.98		
Remote	12	2.15	.73		

Child Concealment					
<i>Demographic Variable</i>	<i>n</i>	<i>M</i>	<i>SD</i>	<i>t</i>	<i>p</i>
Child Gender				.74	.46
Female	31	2.49	.65		
Male	30	2.36	.72		
Medical Specialty				-.54	.59
Dermatology	34	2.47	.71		
Urology	27	2.37	.64		
Other Medical Condition				.39	.70
Yes	18	2.48	.69		
No	43	2.40	.69		
Parent Gender				.33	.74
Female	51	2.44	.70		
Male	10	2.36	.60		
Mode of data collection				.99	.33
Face to face	49	2.47	.69		
Remote	12	2.24	.67		

Child Illness Perceptions					
<i>Demographic Variable</i>	<i>n</i>	<i>M</i>	<i>SD</i>	<i>t</i>	<i>p</i>
Child Gender				-2.29	.03*
Female	31	3.02	.67		
Male	30	3.43	.70		
Medical Specialty				-3.50	.00*

Dermatology	34	2.95	.58		
Urology	27	3.54	.73		
Other Medical Condition					
Yes	18	3.08	.75		
No	43	3.27	.70		
Parent Gender					
Female	51	3.19	.72		
Male	10	3.38	.68		
Mode of data collection					
Face to face	49	3.19	.72		
Remote	12	3.33	.68		

Parent Stigma					
<i>Demographic Variable</i>	<i>n</i>	<i>M</i>	<i>SD</i>	<i>t</i>	<i>p</i>
Child Gender					
Female	31	2.34	.74		
Male	30	2.55	.80		
Medical Specialty					
Dermatology	35	2.51	.74		
Urology	26	2.36	.81		
Other Medical Condition					
Yes	18	2.82	.77		
No	43	2.29	.72		
Parent Gender					
Female	51	2.50	.74		
Male	10	2.16	.90		
Mode of data collection					
Face to face	49	2.48	.77		
Remote	12	2.30	.77		

Note. * $p < .05$, ** $p < .01$

Demographic Variables and Test Variable – Mann Whitney U Test

Child Psychosocial Difficulties					
<i>Demographic Variable</i>	<i>n</i>	<i>Mdn</i>	<i>U</i>	<i>z</i>	<i>p</i>
Gender					
Female	31	5.00			
Male	30	6.00			
Medical Specialty					
Dermatology	34	5.50			
Urology	27	6.00			
Other Medical Condition					
Yes	18	8.00			
No	43	5.00			
Parent Gender					
Female	51	6.00			

Male	10	3.50			
Mode of data collection			255.00	-.71	.48
Face to face	49	6			
Remote	12	4.5			

Parent Concealment					
<i>Demographic Variable</i>	<i>n</i>	<i>Mdn</i>	<i>U</i>	<i>z</i>	<i>p</i>
Child Gender			309.00	-2.26	.02*
Female	31	1.86			
Male	30	2.07			
Medical Specialty			266.50	-2.81	.01**
Dermatology	34	1.86			
Urology	27	2.14			
Other Medical Condition			318.00	-1.20	.27
Yes	18	1.93			
No	43	2.00			
Parent Gender			242.00	-.26	.78
Female	51	2.00			
Male	10	1.93			
Mode of data collection			210.50	-1.52	.13
Face to face	49	2.00			
Remote	12	1.86			

Note. * $p < .05$, ** $p < .01$

Demographic Variables and Test Variable – One-way ANOVAs

Child Stigma					
<i>Demographic Variable</i>	<i>n</i>	<i>M</i>	<i>SD</i>	<i>F</i>	<i>p</i>
Child ethnicity				1.35	.27
White British/ White Other	46	2.42	1.01		
Asian/ Asian British	7	2.81	.46		
Black/ Black British	4	2.13	.57		
Other / Prefer not to say	4	1.69	.51		
School Year				1.17	.60
Year 3-4	7	2.18	.74		
Year 5-6	16	2.21	.72		
Year 7-8	18	2.43	1.2		
Year 9-10	20	2.59	.91		
Time of Onset				1.43	.25
Present at Birth	24	2.14	.66		
Less than 5years old	23	2.54	.88		
5 years or older	14	2.57	1.17		
Hospital Attendance				1.21	.31
1-2 times per year	30	2.21	.87		
3-10 times per year	25	2.61	.93		
More than 10 times per year	6	2.39	1.25		
Medical Condition				2.28	.07

Eczema	19	2.50	.89		
Epidermolysis Bullosa	9	2.38	.81		
Psoriasis	6	3.31	1.02		
Bladder Diagnosis	12	2.20	.69		
Kidney Diagnosis	15	2.06	1.05		
Caregiver age				.28	.76
Up to 39 years	18	2.30	1.32		
40 – 49 years	18	2.48	.80		
50+ years	6	2.15	.51		
Caregiver ethnicity				.95	.42
White British/ White Other	46	2.42	1.01		
Asian/ Asian British	8	2.67	.57		
Black/ Black British	4	2.13	.57		
Other / Prefer not to say	3	1.67	.63		

Child Concealment

<i>Demographic Variable</i>	<i>n</i>	<i>M</i>	<i>SD</i>	<i>F</i>	<i>p</i>
Child ethnicity				.35	.79
White British/ White Other	46	2.39	.74		
Asian/ Asian British	7	2.65	.38		
Black/ Black British	4	2.50	.30		
Other / Prefer not to say	4	2.28	.76		
School Year				.93	.33
Year 3-4	7	2.73	.55		
Year 5-6	16	2.26	.57		
Year 7-8	18	2.51	.69		
Year 9-10	20	2.37	.70		
Time of Onset				.60	.56
Present at Birth	24	2.31	.66		
Less than 5 years old	23	2.47	.68		
5 years or older	14	2.54	.73		
Hospital Attendance				1.17	.32
1-2 times per year	30	2.30	.74		
3-10 times per year	25	2.58	.55		
More than 10 times per year	6	2.43	.68		
Medical Condition				3.09	.02*
Eczema	19	2.27	.65		
Epidermolysis Bullosa	9	2.54	.83		
Psoriasis	6	2.98	.51		
Bladder Diagnosis	12	2.71	.57		
Kidney Diagnosis	15	2.09	.58		
Caregiver age				.15	.86
Up to 39 years	18	2.35	.77		
40 – 49 years	18	2.47	.67		
50+ years	6	2.42	.66		
Caregiver ethnicity				.34	.80
White British/ White Other	46	2.39	.74		

Asian/ Asian British	8	2.63	.36
Black/ Black British	4	2.50	.30
Other / Prefer not to say	3	2.24	.93

Child Illness Perceptions					
Demographic Variable	<i>n</i>	M	SD	F	p
Child ethnicity				1.22	.31
White British/ White Other	46	3.19	.73		
Asian/ Asian British	7	3.10	.42		
Black/ Black British	4	3.13	.55		
Other / Prefer not to say	4	3.86	.87		
School Year				1.17	.60
Year 3-4	7	3.54	.67		
Year 5-6	16	3.33	.57		
Year 7-8	18	3.00	.84		
Year 9-10	20	3.21	.17		
Time of Onset				2.39	.10
Present at Birth	24	3.45	.72		
Less than 5 years old	23	3.10	.57		
5 years or older	14	3.00	.83		
Hospital Attendance				2.07	.14
1-2 times per year	30	3.38	.58		
3-10 times per year	25	3.11	.74		
More than 10 times per year	6	2.83	1.02		
Medical Condition				3.72	.01**
Eczema	19	3.00	.60		
Epidermolysis Bullosa	9	3.02	.36		
Psoriasis	6	2.72	.82		
Bladder Diagnosis	12	3.73	.64		
Kidney Diagnosis	15	3.40	.71		
Caregiver age				.83	.44
Up to 39 years	18	3.03	.87		
40 – 49 years	18	3.34	.62		
50+ years	6	3.31	.70		
Caregiver ethnicity				.49	.69
White British/ White Other	46	3.19			
Asian/ Asian British	8	3.26			
Black/ Black British	4	3.13			
Other / Prefer not to say	3	3.70			

Parent Stigma					
<i>Demographic Variable</i>	<i>n</i>	<i>M</i>	<i>SD</i>	<i>F</i>	<i>p</i>
Child ethnicity				1.92	.31
White British/ White Other	46	2.48	.74		
Asian/ Asian British	7	2.77	.76		
Black/ Black British	4	2.20	.69		
Other / Prefer not to say	4	1.70	.95		

School Year				1.76	.17
Year 3-4	7	1.94	.47		
Year 5-6	16	2.39	.75		
Year 7-8	18	2.42	.87		
Year 9-10	20	2.69	.73		
Time of Onset				.23	.79
Present at Birth	24	2.43	.95		
Less than 5years old	23	2.52	.63		
5 years or older	14	2.34	.69		
Hospital Attendance				2.29	.08
1-2 times per year	30	2.34	.70		
3-10 times per year	25	2.42	.78		
More than 10 times per year	6	3.10	.86		
Medical Condition				1.22	.31
Eczema	19	2.62	.82		
Epidermolysis Bullosa	9	2.40	.71		
Psoriasis	6	2.33	.52		
Bladder Diagnosis	12	2.67	.97		
Kidney Diagnosis	15	2.12	.59		
Caregiver age				.03	.97
Up to 39 years	18	2.41	.65		
40 – 49 years	18	2.47	.85		
50+ years	6	2.40	1.09		
Caregiver ethnicity				.65	.59
White British/ White Other	46	2.48	.74		
Asian/ Asian British	8	2.55	.94		
Black/ Black British	4	2.20	.69		
Other / Prefer not to say	3	1.93	1.01		

Note. * $p < .05$, ** $p < .01$

Demographic Variables and Test Variable – Kruskal Wallis

Child Psychosocial Difficulties				
Demographic Variable	<i>n</i>	<i>Mdn</i>	χ^2	<i>p</i>
Child ethnicity			2.71	.44
White British/ White Other	46	6.00		
Asian/ Asian British	7	7.00		
Black/ Black British	4	4.00		
Other / Prefer not to say	4	3.50		
School Year			6.78	.08
Year 3-4	7	5.00		
Year 5-6	16	4.00		
Year 7-8	18	6.50		
Year 9-10	20	6.00		
Time of Onset			4.04	.13
Present at Birth	24	4.00		
Less than 5 years old	23	6.00		

5 years or older	14	7.50		
Hospital Attendance			8.49	.01*
1-2 times per year	30	4.00		
3-10 times per year	25	7.00		
More than 10 times per year	6	8.50		
Medical Condition			3.01	.56
Eczema	19	6.00		
Epidermolysis Bullosa	9	4.00		
Psoriasis	6	5.50		
Bladder Diagnosis	12	5.50		
Kidney Diagnosis	15	6.00		
Caregiver age			.35	.84
Up to 39 years	18	5.50		
40 – 49 years	18	6.00		
50+ years	6	7.00		
Caregiver ethnicity			1.00	.80
White British/ White Other	46	6.00		
Asian/ Asian British	8	6.50		
Black/ Black British	4	4.00		
Other / Prefer not to say	3	6.00		

Parent Concealment

Demographic Variable	<i>n</i>	Mdn	X ²	<i>p</i>
Child ethnicity			5.15	2.71
White British/ White Other	46	1.86		
Asian/ Asian British	7	2.14		
Black/ Black British	4	2.50		
Other / Prefer not to say	4	1.93		
School Year			3.60	.06
Year 3-4	7	1.57		
Year 5-6	16	2.00		
Year 7-8	18	2.00		
Year 9-10	20	2.00		
Time of Onset			.40	.82
Present at Birth	24	2.00		
Less than 5 years old	23	1.86		
5 years or older	14	1.93		
Hospital Attendance			.73	.70
1-2 times per year	30	1.86		
3-10 times per year	25	2.00		
More than 10 times per year	6	2.00		
Medical Condition			14.04	.01*
Eczema	19	2.00		
Epidermolysis Bullosa	9	1.43		
Psoriasis	6	1.72		
Bladder Diagnosis	12	2.14		
Kidney Diagnosis	15	2.00		

Caregiver age			4.92	.09
Up to 39 years	18	2.00		
40 – 49 years	18	2.14		
50+ years	6	1.93		
Caregiver ethnicity			6.13	.11
White British/ White Other	46	1.86		
Asian/ Asian British	8	2.14		
Black/ Black British	4	2.50		
Other / Prefer not to say	3	1.86		

Note. * $p < .05$, ** $p < .01$

Appendix L: Parametric correlation (Pearsons)

Pearsons Bivariate Correlations between Measures of Stigma, Concealment, Illness Perceptions and Children's Psychosocial Difficulties

Measure	1	2	3	4	5	6
1.Child Stigma Scale	–					
2.Child Secrecy Scale	.606**	–				
3.CATIS	-.685**	-.306*	–			
4.Parent Stigma Scale	.210	.127	-.220	–		
5.Parent Secrecy Scale	.030	-.011	.020	.346**	–	
6.SDQ Internalisation Score	.459**	.275*	-.397**	.386**	.030	–

Note. N = 61 on all measures. * $p < .05$. ** $p < .01$