The Impact of Stigma and Visible Difference on Children & Adolescents Living with Physical Health Conditions

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D.Clin.Psy. Thesis (Volume 1)

2020

University College London

UCL Doctorate in Clinical Psychology

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.

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Overview

This thesis focuses on the impact of stigma and visible difference on Children and Young People (CYP) living with Physical Health Conditions.

Part 1 is a systematic review considering different forms of stigma, concealment and disclosure practices in CYP living with chronic health conditions. It combines the findings of fourteen studies. Findings suggested that CYP experience self-stigma and enacted stigma, with enacted stigma the most prevalent. The impact of how concealable a chronic health condition could be, was frequently reported. Offering interventions which target different forms of stigma and preparing for unplanned disclosures could be an effective way of managing distress in CYP with physical health conditions.

Part 2 is an empirical study considering the impact of self-stigma and visibility on concealment, illness attitudes and psychosocial difficulties in CYP with physical health conditions. Findings suggested that CYP with visible physical health conditions reported poorer illness attitudes and greater psychosocial difficulties than CYP with less visible conditions. Greater self-stigma predicted poorer illness attitudes, greater concealment and more psychosocial difficulties in CYP with physical health conditions. Timely interventions with CYP with visible physical health conditions in particular, could improve the relationship CYP have with their condition and their overall wellbeing. This was a joint project with another University College London (UCL) trainee who investigated psychosocial processes in CYP with physical health conditions and their parents.

Part 3 is a critical appraisal of the systematic review and empirical paper. It includes personal reflections and critiques on the methodology and data collection process.

Impact Statement

Living with a physical health condition in childhood or adolescence can increase the risk of developing psychological difficulties (Gamwell et al., 2018). 50% of mental health problems are established by the age of 14-years-old (Kessler et al., 2005). Understanding more about the factors which influence the relationships Children and Young People (CYP) have towards their physical health condition will allow services to develop informed and effective interventions to reduce distress and improve outcomes for this population. This thesis looked at specific factors which can influence the relationships between CYPs and their physical health condition; with a focus on stigma and visible or less visible differences.

The findings of the systematic review suggested that CYP experience stigma in different forms and report experiences of self and enacted stigma, a distinction not routinely made in research with CYP. This highlights the importance of considering both individualised approaches and wider systemic initiatives to reduce the stigmatisation of CYP with physical health difficulties. CYP discussed different methods, reasons and consequences for attempting to conceal or disclose their physical health condition to others. It could be valuable for clinicians to consider how to support CYP and their families to prepare for and manage the impact of disclosures.

The findings from the empirical paper suggest CYP with visible physical health conditions report poorer illness attitudes and more emotional and peer difficulties than CYP with less visible difference. This is the first study to report these differences and endorses the narrative of heightened appearance related concerns in CYP (Crerand et al., 2017). These findings suggest that it could be important to take account of the impact of visibility and self-stigma when supporting CYP, particularly if they present in distress.

The aim is to disseminate these findings to the clinical and research community by publishing in peer reviewed journals and presenting to paediatric specialists. This could enhance the current literature and raise awareness of the experiences of CYP with physical health conditions.

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Acknowledgments

Most importantly I would like to thank the children, young people and caregivers who participated in this research. I feel hugely privileged that they allowed us a brief insight into their experiences of living with a physical health condition. Having the opportunity to meet them has enriched my understanding and knowledge of physical health conditions beyond the aims of this thesis, and I am in awe of their bravery and resilience.

To my fellow colleague, trainee and friend Claire, thank you being my thesis buddy, for showing me the bright and shiny student centre and for being a huge support through the last few years. I do not know how I would have completed this thesis without you.

To my supervisor Kristina Soon, thank you for all of your support throughout the research project. Thank you for dedicating your time amongst an intensely busy schedule, for being patient and for trying to get the best out of me and the project. I am hugely grateful for having you as a supervisor.

I would also like to express my gratitude to Cat Andrew who navigated Claire and me through complex hospital processes and buildings, and for dedicating time to us and our projects. We are indebted to you and your excel expertise.

I am truly thankful for my family and friends. Particularly my husband who has been unwavering in patience, encouragement and support. Without you, this journey would have been much harder. I would also like to thank my Mum for bringing me over the final hurdle and to my friends for the positivity and motivational snacks. Lastly, to my fellow trainees, the hive minds of FAQs.

Part 1: Literature Review

Stigma, Concealment and Disclosure practices in Children & Adolescents with Chronic Health Conditions: A Systematic Review

Abstract

Aims: Children and Young People (CYP) with chronic health conditions can feel stigmatised because of their condition. Yet, there is limited research exploring in detail the different forms of stigma. Choosing to conceal or disclose a chronic health condition can be complex and affect the safety and wellbeing of CYP. This review aimed to increase the understanding of CYP with a range of chronic health conditions by exploring different forms of stigma and discussing both concealment and disclosure practices.

Method: Embase, MEDLINE and PsycINFO were searched using terms related to CYP, stigma, concealment and disclosure. The search yielded 294 articles, 53 of which were studied for more detailed evaluation. 14 studies were selected including qualitative, quantitative and mixed methods designs, and were critically appraised in detail. A narrative synthesis was used.

Results: Enacted stigma was the most prevalent form of stigma reported amongst CYP. Examples of self-stigma, internalised, perceived and anticipated stigma were also reported. CYP commonly wanted to conceal their condition to avoid self and enacted stigma. Forced disclosure following an unexpected revealing of the condition was frequently reported and could elicit distress. A theme of concealability emerged and that particular features of a condition can make it more or less concealable, which in turn may impact stigma, concealment and disclosure practices.

Conclusion: Chronic health stigma can exist in different forms and having a greater understanding can help to tailor effective clinical interventions accordingly. CYP can engage in both concealment and disclosure processes at the same time and could benefit from support around managing unplanned

disclosures. More research is needed to explore specific relationships between stigma, concealment and disclosure and to explore the impact of concealability.

1. Introduction

Approximately 13-27% of children and young people (CYP) live with chronic physical health conditions (Van Cleave et al., 2010). The literature defines a physical health condition as 'chronic' if the condition is not yet curable, resistant to treatment; and in children, present for at least 3 months before the age of 18 (Jee et al., 2006; Mokkink et al., 2008). Chronic physical health conditions can impact children physically, psychologically and socially (Gamwell et al., 2018; Hysing et al., 2007). Feelings of being different or 'stigmatised' are common experiences reported in children with these conditions (Elstad et al., 2010) with many CYP concealing or disclosing their condition due to the expected or actual responses of others (Benson et al., 2015a).

This review aimed to expand the current understanding of both individual and systemic aspects of stigma, concealment and disclosure which could provide a basis for future research and stigma focused interventions.

1.1 Stigma

Individuals are believed to be stigmatised when they possess behaviour, signs or attributes that are undesired or different to those which society considers 'normal' (Goffman, 2009). When individuals are labelled or stereotyped based on this difference, it can create an 'us and 'them' distinction impacting how individuals see themselves in relation to others and how individuals are treated by others (Link & Phelan, 2001). There are commonalities between stigma and other conceptual models such as prejudice. Stigma models and research are most often concerned with individual-level characteristics, for example, illness or identity difference. By comparison prejudice models often highlight differences between group-level characteristics, for example, gender, race, age and/or class (Phelan et al., 2008; Stuber et al., 2008).

It is possible that experiences of stigma differ across certain illnesses, identities and at different ages and stages of development. The focus of this review is specifically to explore the experiences of stigma in CYP with chronic health conditions.

CYP with chronic health conditions may be at increased risk of stigmatisation based on symptoms or attributes arising from their condition which differentiates them from what is considered normal by society (Dos Santos et al., 2017). Any form of stigma can contribute to psychological distress and adverse outcomes (Mak et al., 2007). However, stigma is multifaceted and can exist in different forms with an individual or system experiencing several forms of stigma concurrently. The types of stigma discussed in this review will be enacted stigma and three forms of self-stigma: anticipated stigma, perceived stigma and internalised stigma. Enacted stigma refers to the experiences of discrimination, prejudice or negative stereotyping from others to a stigmatised individual (Earnshaw & Chaudoir, 2009; Kinsler et al., 2007). Anticipated stigma occurs when an individual expects discrimination, prejudice or negative stereotyping because of a stigmatised identity (Earnshaw et al., 2013; Quinn & Chaudoir, 2009). Perceived stigma occurs when an individual believes that others hold negative judgements, stereotypes or attitudes towards them because of their stigmatised identity (Van Brakel, 2006). Lastly, internalised stigma describes the degree to which individuals accept negative stereotypes and discriminatory beliefs as being true to themselves (Quinn & Earnshaw, 2013; Teh et al., 2014). Although stigma shares conceptual underpinnings with other processes such as prejudice and discrimination, some researchers propose that the psychosocial stress induced by stigma can be distinct from that of discrimination and prejudice. For example, it is suggested that anticipating or perceiving stigma experiences to happen to oneself can be present even when discriminatory treatment is not experienced (Stuber et al., 2008). Other literature and definitions used in the research indicate there is an overlap between discriminatory actions and enacted stigma. Therefore, it is important to be explicit about the different types of stigma for appropriate inferences to be drawn.

There are also many reasons to specifically explore self and enacted stigma in CYP with chronic health conditions. Firstly, childhood and adolescence are critical periods of identity and psychosocial development (MacLeod & Austin, 2003), and feelings of difference or exposure to negative judgements may define how CYP think of themselves and their chronic health condition in adulthood. Secondly, many current studies with samples of CYP and chronic health conditions report stigma as a general concept or broadly comment on feelings of difference but do not differentiate types of stigma. Having a better understanding of different types of stigma in CYP with different chronic health conditions could enable professionals, academics, caregivers and CYP to pursue more effective ways of reducing these stigmas if they are triggering or maintaining distress. For example, individual or systemic psychological support managing specific internalised stigma related cognitions or beliefs can help CYP improve their self-efficacy, self-esteem (Yanos et al., 2011) and reduce the likelihood of mental health difficulties (Austin et al., 2004; Gamwell et al., 2018). Whereas, public awareness campaigns are more likely to target enacted stigma and the beliefs and actions of others (Evans-Lacko et al., 2014; Quinn & Earnshaw, 2013). Lastly, exploring the different forms of stigma in CYP with chronic health conditions will bring the literature up to date with other areas of stigma research which discuss these concepts.

1.2 Concealment and Disclosure

Concealment can be thought of as any conscious behaviour or effort to hide information from others (Allen & Carlson, 2003). Evidence suggests that CYP with chronic health conditions choose to conceal their condition or aspects of it, to avoid stigmatising experiences (Bachmann et al., 2009; Jantzen et al., 2009). Strategies include hiding the condition entirely, masking or passing off symptoms (Williams & Chapman, 2011), hiding side effects of a condition (Mu, 2008), or ensuring medical interventions are kept secret (McEwan et al., 2004). Research suggests that concealing any type of stigmatising identity can negatively impact physical and psychological quality of life (Quinn et al., 2017; Velsor-Friedrich et al., 2004).

Disclosure occurs when information is shared about oneself (Bazarova & Choi, 2014). Many CYP with chronic health conditions face a dilemma when deciding whether to disclose their condition to others and may choose to for reasons of necessity, practicality or in the context of supportive relationships (McMurray et al., 2001; Moola et al., 2011). Disclosure can elicit positive outcomes, for example improving self-confidence (Kaushansky et al., 2017), but it can also lead to discrimination, rejection and bullying from others (Nahal et al., 2019). CYP have been found to employ different strategies to disclosing their chronic health conditions such as indiscriminately disclosing, selectively disclosing, or passively disclosing through visibility of symptoms (Pihlaskari et al., 2019; Werner et al., 2019).

While concealment and disclosure are often used interchangeably, the evidence discussed suggests they may involve different cognitive and motivational processes. Therefore, it is important to separate the two experiences in CYP with chronic health conditions and provide a wider understanding of the strategies, rationale given and possible implications of both concealing and disclosing a chronic health condition.

1.3 Stigma, Concealment and Disclosure

Research suggests that those who have experienced stigma, anticipate stigma occurring or report greater internalised stigma are less likely to disclose a stigmatising chronic health condition to others (Hernandez, 2011; Lee et al., 2017). Research with CYP is very limited and is mostly restricted to CYP with epilepsy. Findings show that a common reason given by CYP for concealing epilepsy is to avoid negative judgement or treatment from others (Barned et al., 2016; Houston et al., 2000). Due to the negative outcomes which can arise from experiencing any form of stigma (Quinn & Chaudoir, 2009; Walker & Reznik, 2014), concealing a condition (Quinn et al., 2017), and receiving negative responses to a disclosure (Nahal et al., 2019), it is important to understand how these experiences relate to each other.

1.4 What this review adds

Currently stigma, concealment and disclosure in CYP with chronic health conditions are not well researched. These constructs are not typically the focus of the studies and the majority of studies are of CYP with epilepsy.

This review aimed to synthesise the existing literature on experiences of stigma, concealment and disclosure in CYP with a range of chronic health conditions. The goal was to provide a better understanding of stigma, concealment and disclosure experiences in relation to different conditions. Uniquely, it will explore different forms of stigma and treat concealment and disclosure as distinct concepts.

1.5 Overall Aims

1. Describe the forms of stigma reported by CYP with chronic health conditions.
2. Discuss any methods, rationale or implications CYP report for both concealing a chronic health condition and disclosing a chronic health condition
3. Explore whether there is a relationship between stigma experienced by CYP and concealment and/or disclosure of chronic health conditions based on the self-reported experiences of CYP.

2. Methods

2.1 Search Strategy

A systematic search of the literature was undertaken by using the following three electronic databases; Embase (2000-present), MEDLINE (2000-present) and PsycINFO (2000-present). There were four search terms: CYP, chronic health conditions, stigma and concealment/disclosure. Both controlled vocabulary and MESH terms were used, along with combining search terms using Boolean ‘Or’ and ‘And’. No limits were applied (see Table 1 for all search terms used). Chronic health condition terms were taken from previous review studies of CYP with chronic health conditions (Bennett et al., 2015; Law et al., 2014; Secinti et al., 2017). Citation and hand searching were also conducted.

Table 1:

Search Terms

Children, Young People	Chronic Physical Health Conditions	Stigma	Concealment and Disclosure
Child* or Children* or teen* or teenager* or young person*' or youth* or youngster* or tween* or adolescent* or kid* or paediatric* or pediatric*	'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 rheumatismor 'sickle cell' or 'spina bifida*' or 'visual impairment' or respiratory or derm* or 'facial difference' or 'chronic pain'	Stigma* or stigmatised or stigmatized	concealable or conceal* or hidden or cover* or disguise* or hide or visible or invisible or disclosure* or disclose or 'keep secret' or 'self-disclose*'

2.2 Study Selection Criteria

Inclusion and exclusion criteria were defined for types of studies, participants and outcomes.

Studies

All research designs were included: quantitative, qualitative and mixed methods studies. Both peer reviewed journals and grey literature were included. 'Grey literature' is defined as dissertations or unpublished studies that are not controlled by commercial publication organisations (Benzies et al., 2006; Turner et al., 2005). Only studies published in English were included due to the limited resources of the reviewer to translate papers. Review papers, books, conference reports or discussion articles were excluded.

Participants

Studies with CYP aged 0-18 of either sex with a chronic physical health condition were included. Papers were excluded if the average age of a sample was above 18- years old, had no related findings to stigma or disclosure, and where experiences of the CYP were reported by another source only (e.g. caregiver, professional etc). Papers were included if CYP reports could be separated from the reports of others (for example, caregiver, professional etc). CYP with AIDS, HIV or HIV-related health illnesses were excluded. The literature suggests that CYP and adults with HIV-related conditions are subject to multiple layers of stigma (Marsicano et al., 2014) which often intersect with ethnic minority status, gender, sexuality (Bogart et al., 2008; Rao et al., 2008; Wolitski et al., 2009) and substance use or addiction (Earnshaw et al., 2015). Although other chronic health conditions may present with aspects of intersectionality such as epilepsy and religion (Bartolini et al., 2011), AIDS and HIV conditions present with a greater range of complex intersections and have therefore been excluded for the purpose of this review.

Study Outcomes

In terms of study outcomes, studies were included that:

- Explicitly examined stigma experiences in CYP with chronic health conditions reported by the CYP as primary focus or as sub-focus of the study

- Explicitly examined concealment or disclosure in CYP with chronic health conditions reported by CYP as primary or as sub-focus of the study

2.3 Relevant Definitions

There are multiple definitions of stigma used across academic research and contemporary literature. This review will define stigma by 1) self-stigma which will include experiences of stigma internal to an individual including internalised stigma, anticipated stigma and perceived stigma and 2) enacted stigma which includes the actual experiences of stigma which are external to the individual. These definitions will help to distinguish the internal and external stigma experiences of CYP (Moore et al., 2013; Quinn & Earnshaw, 2013; Van Brakel, 2006). The definitions used in this review are summarised in Table 2.

Table 2:

Definitions of stigma, concealment and disclosure used in this review

Terms		Definition
Self-Stigma	Internalised Stigma:	When individuals internalise negative judgements, stereotypes and prejudices associated with having a stigmatised identity, trait or characteristic and apply to the self (Quinn & Earnshaw, 2013; Teh et al., 2014)
	Anticipated: Stigma	The degree to which individuals believe that others will stigmatise them i.e., discriminate, prejudice or devalue them base on certain identity, trait or characteristic (Earnshaw et al., 2013; Quinn & Chaudoir, 2009).
	Perceived: Stigma	The belief an individual hold about the attitude of others towards a stigmatising

	identity, trait or characteristic (Van Brakel, 2006).
Enacted Stigma	When individuals feel they have experienced discrimination, prejudice or negative stereotyping towards them from others based on a certain identity, trait or characteristic (Earnshaw & Chaudoir, 2009; Kinsler et al., 2007).
Concealment	Can be defined as conscious behaviours or efforts made to decrease the visibility of any limitations (Allen & Carlson, 2003).
Disclosure	Personal information revealed about oneself, typically verbally (Chaudoir & Fisher, 2010) but could also be through other media such as online platforms or social media (Bazarova & Choi, 2014).

2.4 Methods of review

Using the defined selection criteria, a two-stage screening approach identified eligible studies. Stage one included screening the titles and abstracts of all retrieved material from the electronic databases used. Relevant studies progressed to stage two. Stage two involved retrieval of the full text of all studies deemed eligible and were read by the author who applied the inclusion/exclusion criteria to reach the final selection of studies. The reasons for excluding studies is presented in Figure 1. The rationale for excluded studies is provided at each stage. Fourteen studies that met all selection criteria were included in the review. This included nine qualitative, three quantitative and two mixed-methods studies.

2.5 Data Extraction and Data Synthesis

The following descriptive information was extracted for each study: author, year, country, overall study aims, study design, data collection (including any questionnaire measures if applicable), chronic health condition, sample characteristics, findings related to type of stigma (as defined in Table 2), whether the study focused on concealment, disclosure or both (Table 3). To capture key findings relating to the aims of this review, the following data was extracted: how stigma, concealment and/or disclosure findings emerged, experiences of each type of stigma, experiences of concealment, experiences of disclosure, experiences of both concealment and disclosure (Table 4). Key findings were identified through a) primary study aims as specified by the author(s) of the paper, b) secondary themes discussed and identified by the author(s) of the paper, c) themes/experiences reported by CYP/author(s) in the papers which the reviewer identified based on the definitions of stigma, concealment and disclosure named in Table 2. Themes that emerged within each of the study aims were also identified by the reviewer (Table 8). The data was synthesised narratively as this method can be used to combine several research designs (Barnett-Page & Thomas, 2009; Lucas et al., 2007). Several review papers looking at similar areas of interest and using a narrative synthesis to combine qualitative, quantitative and mixed method designs were used for guidance (Benson et al., 2015; Lambert & Keogh, 2015). As such, findings from qualitative, quantitative and mixed method designs were combined in relation to the study aims and themes identified.

Figure 1:

PRISMA Flow Diagram

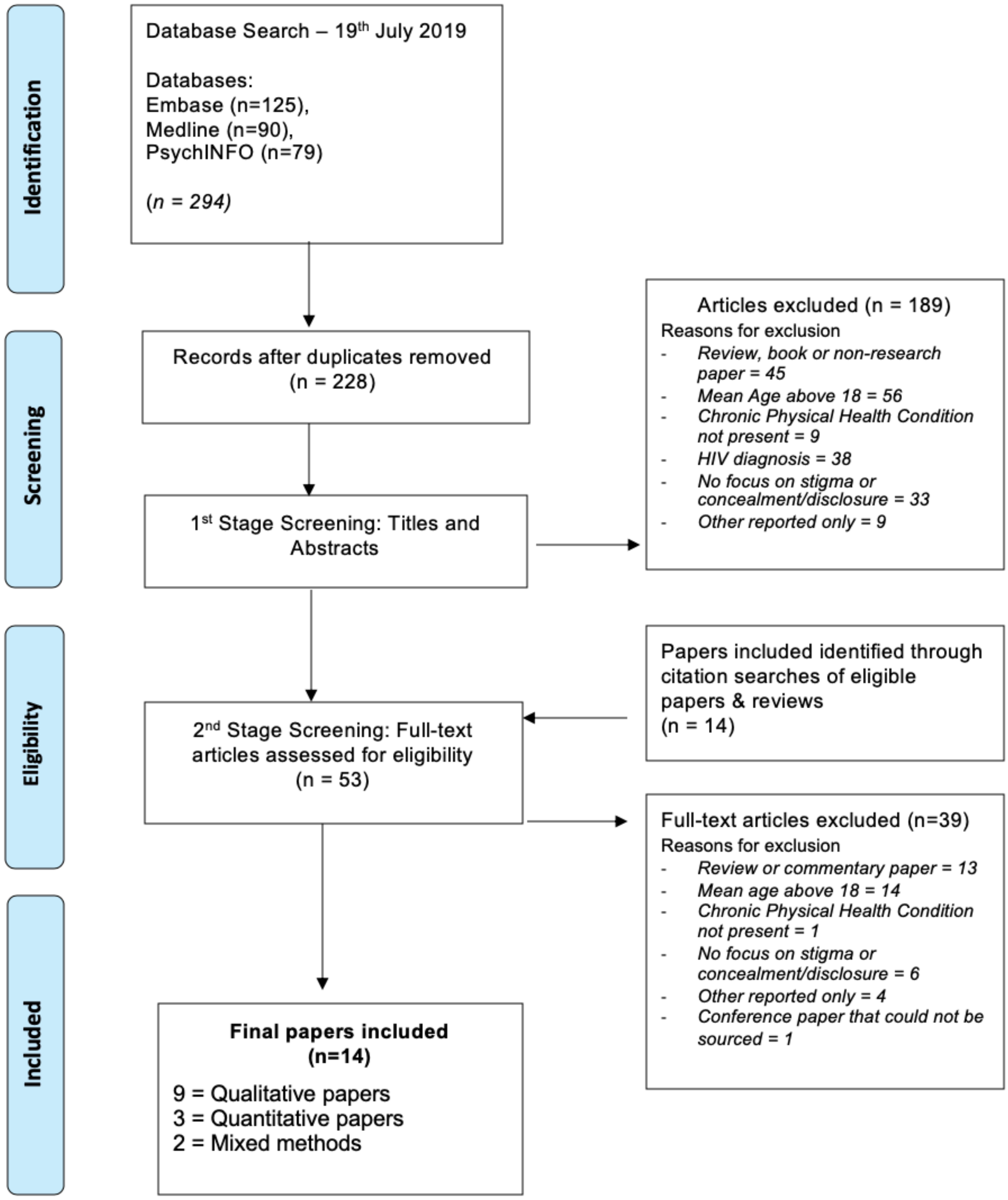

Table 3:

Methodological details and key areas of included studies

Authors *(Country)*	Aims	Study Design	Data Collection	Condition	Sample	Type of Stigma	Concealment or Disclosure or both
Qualitative Studies							
Benson et al., 2015 *Ireland*	Challenges of CYP when disclosing an epilepsy	Exploratory Qualitative *NS*	Semi-structured Questionnaire	Epilepsy	N= 29 Female - 12, Male - 17 MAge = 11y Ethnicity – NR	Internalised Stigma Anticipated Stigma Perceived stigma Enacted Stigma	Both
Kirk & Hinton, 2019 *UK*	How CYP respond to a CHC diagnosis	Exploratory Qualitative *Constructive Grounded Theory*	Semi-Structured Interviews	Multiple Sclerosis	N= 21 Female – 15 Male – 6 Mage = 15y Ethnicity; White =17, South Asian =4	Anticipated Stigma Perceived Stigma Enacted Stigma	Both

Lewis & Parsons, 2008 *UK*	Perceived impact of epilepsy on CYP daily life	Exploratory Qualitative *NS*	Semi-Structured Interviews Open-ended E-surveys	Epilepsy	*Interviews* N=22; Female – 13 Male – 9 Ethnicity; White British=18 British Asian= 3 Mixed Heritage=1 MAge= 12y *Survey* N=44 MAges = 7.3y & 16.9y Ethnicity – NR	Internalised Stigma	Both
Macleod, 2009 *USA*	Everyday lives of adolescent girls with epilepsy	Exploratory Qualitative *Phenomenology approach*	Semi-structured interviews	Epilepsy	N= 4 All Female MAge= 16y Ethnicity - NR	Internalised Stigma Anticipated Stigma Perceived Stigma Enacted Stigma	Both

Moffat et al., 2009 *UK*	General Impact of epilepsy on CYP	Exploratory Qualitative *Grounded theory*	Focus groups and two Semi-Structured Interviews,	Epilepsy	N=22 Female – 11 Male – 11 MAge= 9.6y Ethnicity - NR	Internalised Stigma Anticipated Stigma Perceived Stigma Enacted Stigma	Both
Olsson et al., 2009 *Sweden*	Experience of CYP with celiac disease and gluten-free diet	Exploratory Qualitative *NS*	Focus groups	Celiac Disease	N=47 Females – 32 Male – 15 Aged = 15-18y Ethnicity – NR	Internalised Stigma Anticipated Stigma Perceived Stigma Enacted Stigma	Both
Rhee, et al., 2007 *USA*	Experiences and coping strategies of CYP with asthma	Exploratory Qualitative *NS*	Focus Groups	Asthma	N=19 Female – 11 Male – 8 MAge= 13.4y & 16.5y Ethnicity - NR	Internalised Stigma Perceived Stigma Enacted Stigma	Both

Tiemens, et al., 2013 *Canada*	Experience of adolescent girls with a facial difference	Exploratory Qualitative *Phenomenological Approach*	Semi-structured interviews	Cleft Lip & Cleft Palate (Orofacial clefts)	N=7 All Female MAge= 17y Ethnicity; Caucasian=5 Minority =2	Internalised Stigma Anticipated Stigma Perceived Stigma Enacted Stigma	Concealment
Wo et al, 2018, *Malaysia*	Explore experiences and challenges of parents and CYP	Exploratory Qualitative *Phenomenological Approach*	Semi-Structured interviews	Epilepsy	N=15 Female – 7 Male – 8 MAge=12.7y Ethnicity – NR	Anticipated Stigma Perceived Stigma Enacted Stigma	Both
Quantitative Studies							
Krüger et al., 2014 *Finland*	Quality of life, behavioural patterns, experiences of CYP with vitiligo	Cross-Sectional Quantitative	Survey *-Children's Dermatology Life Quality Index - 22-item Questionnaire*	Vitiligo	N=74 Female – 50 Male – 24 Aged = 9.1-14.5y Ethnicity - NR	Enacted Stigma	Concealment

			developed by the authors				
Masnari et al., 2012 *Switzerland*	Stigma in CYP with facial conditions compared to non-disabled peers	Cross-Sectional Quantitative	Survey *-Perceived Stigmatized Questionnaire* *-Parent report size/location of facial difference*	Facial Differences	N= 87 Female – 41 Male – 46 MAge = 6.17y Ethnicity - NR	Enacted Stigma	Concealment
Usitalo, 2002 *USA*	Explore perceived stigma in CYP	Cross-Sectional Quantitative	Survey *-Stigma - Social Perception Questionnaire* *- Physical appearance – Self-Description Questionnaire*	Cranio-Facial Condition	N=83 Female – 30 Male – 53 MAge= 14.6y Ethnicity; Caucasian = 68 African American = 6 Hispanic = 6 Other = 3	Enacted Stigma	Concealment

Mixed Method Studies

Benson et al., 2016 *Ireland*	Experience of stigma, R* between CYP stigma perceptions, seizure variables & communication	Mixed-Methods Study	Semi-Structured Interviews & Survey *-Child Stigma Scale (CSS)* *-Epilepsy Disclosure Scale* *-Talking to Parents about Epilepsy Affect Scale*	Epilepsy	*Interviews* N=33 Female – 20 Male – 13 MAge =11.14y Ethnicity - NR *Survey* N=47 Female – 25, Male – 22 MAge =13.19y Ethnicity - NR	*Qualitative* Internalised Stigma Enacted Stigma *Quantitative* Internalised Stigma	*Qualitative* Concealment *Quantitative* Concealment

Dyson et al., 2010 *England*	Explore pattern of disclosure and reasons for and against disclosure	Mixed-Methods Study	Semi-Structured Interviews & Survey *Author developed survey (Results reported and interpreted together)*	Sickle Cell Disease	*Interviews* N=40 Female – 21, Male – 19 75% Aged 11-18 Ethnicity; Black African = 60%, Black Caribbean = 37.5%, Not stated 2.5% *Survey* N=569 Female – 288, Male – 281 91.6% aged 5-18; Ethnicity; Black African = 60.3%, Black Caribbean = 28.5%, Black Other = 8.1%, Not stated = 3.2%	*Qualitative* Anticipated Stigma Perceived Stigma Enacted Stigma *Quantitative* Enacted Stigma	*Qualitative* Disclosure *Quantitative* Disclosure

N = sample size, MAge – Mean age of sample, y = years old, NS – not specified, CYP – children and young people, R=relationship [a] = communication*

Table 4:

Key Findings: Experiences of stigma, concealment and/or disclosure

Author (year) *Condition* How findings emerged	Self-Stigma			Enacted stigma	Concealment	Disclosure	Stigma and concealment
	Internalised stigma	Anticipated stigma	Perceived stigma				
Qualitative Studies							-
Benson, et al., 2015 *Epilepsy* Key themes emerged from interviews	General Theme* 'Feeling of Differentness' because of Epilepsy': Feeling different to peers	General theme* 'Anticipated Negative Responses to Epilepsy': Worries of being treated differently, being bullied, excluded or pitied.	General Theme* 'Others Perception': Perceived others thinking of them as different weird, and contagious.	General theme* 'Actual Negative Response of Others': Being avoided, teased, bullied by peers.	Concealing epilepsy outside of nuclear family Concealed epilepsy to avoid feeling different, being treated differently	General theme* 'Selective Disclosure to Family': Negative experiences impacted disclosure decisions Disclosed for necessity	-

Kirk & Hinton, 2019 *MS* Key Themes emerged from Interviews	-	Fears of being treated differently	Fear of being defined as different by peers Thought others held beliefs about their physical abilities	Bullied and teased	Concealed MS to avoid distressing responses from others and being asked questions	Selectively disclosed MS to close peers Disclosed MS for necessity and practical support.	-
Lewis & Parsons, 2008 *Epilepsy* Key themes emerged from interviews and e-surveys	Reported feeling not normal or embarrassed because of epilepsy	-	-	-	Concealed condition to avoid embarrassment. Wanted to disclose but feared information would spread to others	Disclosed to close friends Forced to disclose due to onset of symptoms Disclosure reported to enhance peer support and reassurance	-

Macleod, 2009 *Epilepsy* Key themes emerged from interviews and e-surveys	Four CYP reported epilepsy made them different One CYP reported that medication made her feel different Two CYP reported others possess different traits	Worries of being treated differently	-	Reported being bullied, teased and physical violence.	Desire to have more control over concealing condition Avoided conversations about epilepsy Concealed to avoid distressing others	Reported disclosure management strategies Disclosed due to practicality and necessity Disclosure gained peer support. Forced disclosure due to onset of symptoms	-

Moffat et al., 2009 *Epilepsy* Key themes emerged from focus-groups and interviews	Sub-theme* 'Bad Things About Epilepsy:' Feelings of being different, embarrassed & lonely Sub-Theme*: 'Good Things': Normal to feel different	Sub-Theme* 'What Other People Think and Do': Reports of being stared up	Sub-Theme* 'What Other People Think and Do': Perception that others think they are weird	Sub-Theme* 'What Other People Think and Do': Bullied, teased, laughed at, excluded and talked about	Sub-Theme* 'Telling people'*:* Keeping epilepsy a secret Worried others would shared epilepsy diagnosis	Sub-Theme* 'Telling people': Disclosed to cose friends Embarrassed after forced disclosure Disclosed for practical safety Some peers/teachers supportive after disclosure	-

Olsson et al., 2009 *Celiac Disease* Key themes emerged from focus-groups	Sub theme* 'Feelings of Social Deviance': Not feeling normal General theme* 'Being Centre of Attention': GF diet led to feelings of difference	Sub theme* 'Feelings of Social Deviance': Others staring, being pitied	Sub theme* 'Feelings of Social Deviance': Others thinking badly due to food restrictions	General theme* 'Invisible Problem Made Visible': Others staring, making comments and jokes General theme* 'Being Centre of Attention': Others minimising, amplifying, ignoring symptoms	General theme* 'Facing Dietary Deviance': Avoided conversation of GF diet Concealed condition to avoid embarrassment, being pitied General theme* 'Being Centre of Attention': Concealed to avoiding difficult response of others	General theme* 'Facing Dietary Deviance': Disclosure could be helpful

Rhee et al., 2007 *Asthma* Key themes emerged from focus-groups	General Theme* 'Emotional Responses to Living with Asthma': Embarrassed about physical limitations	-	General Theme* 'Perceptions and Attitudes of Others in Response to Teens' Asthma': Perception that others became frustrated with them	General Theme* 'Emotional Responses to Living with Asthma': Ridiculed and, restricted in activities. General Theme* 'Perceptions and Attitudes of Others in Response to Teens' Asthma': Others minimised or amplified symptoms	General Theme* 'Coping Strategies': Masking or disregarding symptoms	General Theme* 'Perceptions and Attitudes of Others in Response to Teens' Asthma': Symptoms misunderstood after disclosure Disclosure could change behaviour of others	-

Tiemens et al., 2013 *Orofacial clefts* Key themes emerged from interviews	General Theme* ‘Struggle with Stigma’: Reported feeling different, ugly. Desire to be normal and accept self	General Theme* ‘Struggle with Stigma’: Perceived not being given same opportunity-es as others, Condition impact friendships and relationships	General Theme* ‘Struggle with Stigma’: Feelings of being devalued by others, not accepted	General Theme* ‘Struggle with Stigma’: Stared at, teased and excluded	Concealing face	-	-
Wo et al., 2018 *Epilepsy*	-	Sub theme* ‘Management of Epilepsy Care’:	Sub theme* ‘Management of Epilepsy Care’:	Sub theme* ‘Impact on Epilepsy on Children’:	Majority of CYP concealed condition	Sub theme* ‘Management of Epilepsy Care’: Selective disclosure to trusted others	-

Key themes emerged from interviews	Not wanting to be treated differently	Viewed as different and weird	Bullied, teased, socially isolated		Forced disclosure increased stigma Disclosure led to emotional support	
Quantitative Studies						
Kruger et al., 2014 *Vitiligo* Sub focus of larger study through author developed questionnaire	-	-	93.7% experienced others asking questions about vitiligo 44.6% reported being picked/nasty comments 21.7% reported being bullied 50% of CYP had been picked on, and teased,	24.4% hid vitiligo 75.6% never hid vitiligo 29.7% avoided situations vitiligo noticed		Sig* R* between being picked on and avoidance behaviours (p<0.01)

Masnari et al., 2012 *Facial conditions* Stigma focus of the study, through validated questionnaire	-	-	-	55.1% reported others were startled by their appearance 82% reported being pitied 62.1% reported others stared 41.4% reported being called names 13.8% reported being laughed at 17.2% reported being bullied 24.1% teased	-	-	CYP reported sig* higher stigma than peers without facial difference Size of the facial different was sig* predictor of stigmatising experiences CYP with facial difference, 25% + of their face reported sig* greater stigma

Usitalo 2002 *Cranio-Facial Condition* Stigma focus of the study, through validated questionnaire				83% reported stigma in social interactions 55.4% reported low stigmatising experiences 44.5% CYP reported moderate or high stigmatising experiences	Sig* relationship between stigmatising experiences and physical appearance Lower satisfaction with physical appearance higher stigmatisation experiences		
Mixed Method Studies							
Bensont et al., 2016 *Epilepsy* Stigma main focus of	Quali* General Theme* 'Internalised Negative Feelings			Quali* General Theme* 'Social Exclusion': Reported exclusion from	Quali* General Theme* 'Concealment': Actively concealed epilepsy		Quant* Sig * R* between self-stigma and concealment = greater self-

study, explored using interviews and validated questionnaire	Towards Epilepsy’: Feeling different or odd General Theme* ‘Teasing/ Bullied’: Feeling embarrassed and ashamed Quant* Neutral perceptions of epilepsy related stigma	events and activities General Theme* ‘Teasing/ Bullied’: Excluded by peers and adults Bullied and teased	stigma score, greater epilepsy concealment score

Dyson et al., 2010 _SCD*_ Disclosure patterns main focus of study, using interviews and author developed survey		Quali*Quant Fear of being excluded	Quali*Quant Perception of being thought of as lazy	Quali*Quant Called lazy by peers 45.8% denied water 57.4% denied toilet breaks 36.3% forced into unsuitable exercise		Quali*Quant 10% reported 1+ person in school did not know 13 reported no bad experiences when peers knew 19 reported bad experiences when peers knew 34 reported no bad experiences when reported to school 17 reported bad experiences when reported to school Disclosed for practical support	Quali*Quant Some CYP reported disclosure, trigged bullying discrimination

CYP = children and young people, General Theme = general theme reported in study, Sub-theme* = sub theme reported in study, Sig* = significant, R*= relationship, Quali* = qualitative findings, Quant* = quantitative findings, Quali*Quant = qualitative and quantitative findings reported together in study*

2.6 Quality Assessment

Two tools were used to critically appraise the studies selected and to guide the strength of the findings discussed in each study. No studies were excluded following the quality assessment. The author appraised all 14 studies using the following tools with a second-rater appraising eight out of 14 studies with a representative proportion of study designs.

2.6.1 Qualitative and Quantitative Quality Assessment

The quality of the quantitative and qualitative studies was appraised using the Standard Quality Assessment Criteria developed by Kmet et al. (2004). This measure was chosen because it offered ways to appraise qualitative and quantitative designs and gave a score that allowed the quality of the studies to be compared. The criteria for evaluating the qualitative papers comprised ten questions assessing: the study design, context, theoretical/model or framework used, sample strategy, data collection and analysis, credibility procedures, conclusions drawn and reflexivity (Appendix A). For the quantitative tool, fourteen questions assessed the study design, method of sample selection, sample characteristics, definition of outcome, sample size, data analysis/reporting, the consideration of variance and confounds, and the conclusions drawn (Appendix B). Each question on the tool was scored depending on the degree to which the specific criteria were met (“yes” = 2, “partial” = 1, “no” = 0). Items that were not applicable to a particular study design were marked “n/a” and excluded from the total score. A summary score was calculated for each paper by summing the total score obtained across relevant items and dividing by the total possible score. Each tool yielded a score between 0-1.0 with higher scores indicating more robust methodology.

2.6.2 Mixed Methods Quality Assessment

The quality of the mixed methods studies were evaluated in two stages. Firstly, using Kmet et al. (2004) Standard Quality Assessment Criteria to evaluate the qualitative and quantitative methodology separately. Secondly, applying the specific criteria relating to mixed methods; the Mixed Methods Appraisal Tool (MMAT) developed by Hong et al. (2018). This framework used the following five questions: 1) Is there an adequate rationale for using a mixed methods design to address the research question? 2) Are the different components of the study effectively integrated to answer the research question? 3) Are the outputs of the integration of qualitative and quantitative components adequately interpreted? 4) Are the divergences and inconsistencies between quantitative and qualitative results adequately addressed? 5) Do the different components of the study adhere to the quality criteria of each tradition of the methods involved? (Appendix C). The final question relies upon the qualitative and quantitative components of the study to be appraised individually for their robustness, hence the rationale for employing Kmet et al. (2004) criteria first. The MMAT does not produce a score but both mixed methods studies in this review were allocated a score for each qualitative and quantitative component.

3. Results

3.1 Quality Appraisal of the Included Studies

Using the Standard Quality Assessment Criteria's (Kmet et al., 2004) the quality of the nine qualitative studies (Table 4) and the three quantitative studies (Table 5) were analysed. From a total score of 0 – 1.0 with 1.0 indicating extremely robust methodolgy, the studies appraised obtained scores ranging from 0.60 – 1.0. The two mixed methods studies (Benson et al., 2016b;

Dyson et al., 2010) were appraised using the Standard Quality Assessment Criteria to determine the quality of the quantitative and the qualitative methodology and then overall as a mixed-methods design using the MMAT (Hong et al., 2018). The average of both quantitative and qualitative methodology combined of these scores were 0.77 (Dyson et al., 2010) and 0.85 (Benson et al., 2016b) when scored using the Standard Quality Assessment and rated overall using the MMAT (Table 6). The second-rater used the critical appraisal tools to rate eight of the papers and a Cohen's kappa was found of κ=0.65, suggesting substantial agreement in ratings (McHugh, 2012). Studies with a score of <0.75 were deemed to be of lower quality and their findings presented cautiously in the narrative synthesis unless similar findings were observed in studies with higher methodological quality. Only two studies were deemed to be of distinctly lower quality than the other twelve; Lewis and Parsons (2008) and Dyson et al. (2010).

Table 5:

Quality Appraisal (Kmet et al., 2004) – Qualitative Study Design

Author	**1.**	**2.**	**3.**	**4.**	**5.**	**6.**	**7.**	**8.**	**9.**	**10.**	**Total (0-20/20)**
Benson et al., (2015)	2	2	2	0	1	2	2	2	2	0	15 (0.75)
Kirk & Hinton, (2019)	2	2	2	2	2	2	2	2	2	2	20 (1.0)
Lewis & Parsons, (2008)	1	0	2	1	1	2	2	0	2	1	12 (0.60)
Macleod (2009)	2	2	2	2	2	2	1	2	2	2	19 (0.95)
Moffat et al., (2009)	2	2	2	2	2	2	2	2	2	1	19 (0.95)
Olsson et al., (2009)	2	2	2	2	2	2	2	2	2	0	18 (0.90)
Rhee et al., (2007)	2	2	2	2	2	2	2	2	2	0	18 (0.90)
Tiemens et al., (2013)	2	2	2	2	2	2	2	2	2	0	18 (0.90)
Wo et al., 2018)	2	2	2	2	2	2	2	0	2	1	17 (0.85)

Questions: 1. Objectives sufficiently described? 2. Study design evident & appropriate? 3. Context for the study clear? Connection to theoretical framework or body of knowledge? 5. Sampling strategy described relevant and justified? 6. Data collection methods described and systematic? 7. Data Analysis clearly described and systematic? 8. Use of verification procedures for credibility? 9. Conclusions supported by the results? 10. Reflexivity of the Account? (All scored 0-2) k

Table 6:

Quality Appraisal (Kmet et al., 2004) Quantitative Study Design

Author	1.	2.	3.	4.	8.	9.	10.	11.	12.	13.	14.	Total (0-22/22)
Kruger et al., (2014)	2	2	1	2	1	2	2	1	1	2	2	18 (0.81)
Masnari et al., (2012)	2	2	2	2	2	2	2	2	2	2	2	22 (1.0)
Usitalo, (2002)	2	2	2	2	2	2	2	2	1	2	2	21 (0.95)

Questions, 1. Objectives sufficiently described? 2. Study design evident & appropriate? 3. Method of selection or source of information described and appropriate?4. Subject characteristics sufficiently described? 8. Outcome clearly defined and robust to measurement bias, means of assessment reported? 9. Sample size appropriate? 10. Analytical methods described and justified & appropriate? 11. Some estimate of variance reported? 12. Controlled for confounding? 13. Results reported in sufficient detail? 14. Conclusions supported by the results? (All scored 0-2) (Question 5,6,7 n/a)

Table 7: *Quality Appraisal MMAT (Hong et al., 2018) Mixed Methods Study Design*
Quality Appraisal MMAT (Hong et al., 2018) Mixed Methods Study Design

Author	Score Qualitative (Kmet)	Score Quantitative (Kmet)	1.	2.	3.	4	5
Benson et al., (2016)	0.75	0.95	Yes	Yes	Yes	Can’t tell	Yes
Dyson et al., (2010)	0.85	0.73	Yes	Yes	Yes	Can’t tell	Can’t tell

Question: 1 Is there adequate rationale for using a mixed methods design to address the research question? 2. Are the different components of the study effectively integrated to answer the research question? 3. Are the outputs of the integration of qualitative and quantitative components adequately interpreted? 4.Are divergences and inconsistencies between quantitative and qualitative results adequately addressed? 5.Do the different components of the study adhere to the quality of criteria of each tradition of the methods involved? (All answers Yes, No, Can’t tell)

3.2 Overview of the Studies Included

In total, 14 studies were included in the review which described the experiences of stigma, concealment and/or disclosure in CYP with chronic health conditions. Seven studies were acquired through the citation searching of relevant topic papers. The studies are mixed in their study designs with nine qualitative, three quantitative and two mixed methods papers. Qualitative studies used different methods to explore and elicit the experiences of CYP, all quantitative studies were cross-sectional, and questionnaire based. No intervention studies were found.

A total of 1,163 CYP were included across the fourteen studies, 53% of the sample were female and 47% male. The average age of CYP across the studies ranged from 4 – 17 years old. The majority of studies were conducted in North American and Europe with three in the US (Usitalo, 2002; Rhee, et al., 2007; MacLeod, 2009), one in Canada (Tiemens et al., 2013), six in the UK and Ireland (Lewis & Parsons, 2008; Moffat et al., 2009; Dyson et al., 2010; Benson et al., 2015; Benson et al., 2016; Kirk & Hinton, 2019), one in Switzerland (Masnari et al., 2012), one in Finland (Krüger, Panske, & Schallreuter, 2014), one in Sweden (Olsson, et al., 2009) and one in Malaysia (Wo et al., 2018). Of the 14 studies, six recruited CYP with epilepsy (Lewis & Parsons, 2008; MacLeod, 2009; Moffat et al., 2009; Benson et al., 2015; Benson et al., 2016; Wo et al., 2018), three recruited CYP with craniofacial conditions, for example cleft lip and cleft palate (Usitalo, 2002; Masnari et al., 2012; Tiemens et al., 2013) and the remaining five studies recruited CYP with asthma (Rhee et al., 2007), celiac disease (Olsson et al., 2009), sickle cell disease (Dyson et al., 2010), multiple sclerosis (Kirk & Hinton, 2019) and vitiligo (Krüger et al., 2014). More detailed sample characteristics, methodological information and key findings from each study relating to the aims of this review are presented in Table 3 and Table 4 above.

Strengths

Twelve out of the 14 studies were found to have high ratings by the quality appraisal tools which suggests that the majority of methodological properties of the studies included were robust (Benson et al., 2015; Benson et al., 2016; Kirk & Hinton, 2019; Krüger et al., 2014; MacLeod, 2009; Masnari et al., 2012; Moffat et al., 2009; Olsson et al., 2009; Rhee et al., 2007; Tiemens et al., 2013; Usitalo, 2002; Wo et al., 2018). Of the studies included, 53% of the samples were females and 47% male and overall CYP were aged between 4 – 17-years old. This suggests that the studies are representative of both male and female CYP and that experiences are captured across several ages and stages of development.

Limitations

The questions on the quality appraisal tools guided the reviewer to critically appraise the studies identified but there were a number of limitations the Standard Quality Assessment Criteria (Kmet et al., 2004) and the MMAT (Hong et al., 2018) did not address. Although the majority of studies described and justified their sampling methods, all 14 studies included used self-selected samples, and four studies required participants to initiate contact with the researcher (Benson et al., 2015; Benson et al., 2016; Kirk & Hinton, 2019; Rhee et al., 2007). Firstly, this suggests that CYP and their families who participated in the research may have had a particular interest in this study area, aims or objectives. Secondly, it could suggest that the samples were biased towards those who have the resources and motivation to participate. Only four studies commented on those who chose not to participate (Lewis & Parsons, 2008; Macleod, 2009; Tiemens, et al., 2013) and only one study, carried out in Malaysia, made reference to adapted materials for several languages (Wo et al., 2018). Evidence suggests that logistical difficulties, language barriers and travel are common barriers to recruitment (Newington & Metcalfe, 2014). Therefore, these are particular samples and may be influenced by the factors and steps involved in participation.

Additionally, only one study made reference to the intersection between stigma and race (Dyson et al., 2010). It is possible that the way in which chronic health conditions are perceived could differ among some communities and cultures (Bartolini et al., 2011) and may trigger more or less stigmatising experiences overall. Considering the growing prevalence of chronic health conditions in the UK and worldwide (Perrin et al., 2014) it is important to note that experiences of stigma might be influenced by other aspects of intersectionality.

3.3 Findings

Findings pertaining to experiences of stigma, concealment and/or disclosure are summarised in Table 4. These findings were then compiled by the reviewer who then identified themes associated with each of the study aims. Key themes identified by the reviewer in relation to each of the study aims are presented in Table 8 with reference to how frequently each theme occurs. Due to the nature of qualitative studies in particular, the exact number of CYP in each study who reported certain themes was not described. Therefore, findings pertaining to stigma, concealment and/or disclosure are presented narratively, combining the themes/results from each of the 14 studies and across qualitative, quantitative and mixed method designs.

Table 8: Theme count from included studies

Theme count from included studies

Theme	Frequency of Occurrence	Relevant References
Self-Stigma		
Internalised Stigma	9	Benson et al., 2015; Benson et al., 2016; Kruger et al., 2014; Lewis & Parsons, 2008; Macleod, 2009; Moffat et al., 2009; Olsson et al., 2009; Rhee et al., 2007; Tiemens et al., 2013.
Anticipated Stigma	8	Benson et al., 2015; Dyson et al., 2010; Kirk & Hinton, 2019; Macleod, 2009; Moffat et al., 2009; Olsson et al., 2009; Tiemens et al., 2013; Wo et al., 2018.
Perceived Stigma	8	Benson et al., 2015, Dyson et al., 2010; Kirk & Hinton, 2019; Moffat et al., 2009; Olsson et al., 2009; Rhee et al., 2007; Tiemens et al., 2013; Wo et al., 2018.
Enacted Stigma	13	Benson et al., 2015; Benson et al., 2016; Dyson et al., 2010; Kirk & Hinton, 2019; Kruger et al., 2014; Macleod, 2009; Masnari et al., 2012; Moffat et al., 2009; Olsson et al., 2009; Rhee et al., 2007; Tiemens et al., 2013; Usitalo, 2002; Wo et al., 2018
Concealment Practices	9	Benson et al., 2015; Benson et al., 2016; Dyson et al., 2010; Kruger et al., 2014; Macleod, 2009; Moffat et

		al., 2009; Olsson et al., 2009; Tiemens et al., 2013; Wo et al., 2018.
Methods of Concealment	4	Kruger et al., 2014; Macleod, 2009; Olsson et al., 2009; Rhee et al., 2007,
Rationale for Concealment	6	Benson et al., 2015; Kirk & Hinton, 2019; Lewis & Parsons, 2008; Macleod, 2009; Moffat et al., 2009; Olsson et al., 2009.
Selective Disclosure Practice	7	Benson et al., 2015; Dyson et al., 2010; Kirk & Hinton, 2019; Lewis & Parsons, 2008; Macleod, 2009; Moffat et al., 2009; Wo et al., 2018
Forced Disclosure	9	Benson et al., 2015; Kirk & Hinton, 2019; Lewis & Parsons, 2008; Macleod, 2009; Moffat et al., 2009; Olsson et al., 2009; Rhee et al., 2007; Wo et al., 2018.
Rationale for Disclosure	6	Benson et al., 2015; Dyson et al., 2010; Kirk & Hinton, 2019; Lewis & Parsons, 2008; Macleod, 2009; Moffat et al., 2009.
Advantages to Making a Disclosure	7	Dyson et al., 2010; Kirk & Hinton, 2019; Lewis & Parsons, 2008. Macleod, 2009; Olsson et al., 2009; Rhee et al., 2007; Wo et al., 2018.
Disadvantages to Making a Disclosure	4	Dyson et al., 2010; Macleod, 2009; Olsson et al., 2009; Rhee et al., 2007.
Relationship between Stigma and Concealment	7	Benson et al., 2015; Benson et al., 2016; Dyson et al., 2010; Kirk & Hinton, 2019; Kruger et al., 2014,

		Macleod, 2009; Rhee et al., 2007; Wo et al., 2018.
Relationship between Stigma and Disclosure	8	Benson et al., 2015, Kirk & Hinton, 2019, Lewis & Parsons, 2008, Macleod 2009, Moffat et al., 2009, Olsson et al., 2009, Rhee et al., 2007, Woo et al., 2018
Concealability	11	Benson et al., 2015; Benson et al., 2016; Kirk & Hinton, 2019; Lewis & Parsons, 2010; Macleod, 2009, Masnari et al., 2012; Olsson et al., 2009; Rhee et al., 2007; Tiemens et al., 2013; Usitalo, 2002, Wo et al., 2018.

3.3.1 Self-Stigma

Internalised Stigma

Internalised stigma was reported in nine out of 14 studies (Benson et al., 2015; Benson et al., 2016; Kruger et al., 2014; Lewis & Parsons, 2008; Macleod, 2009; Moffat et al., 2009; Olsson et al., 2009; Rhee et al., 2007; Tiemens et al., 2013). Examples included CYP holding negative beliefs about themselves, feeling embarrassed, labelling themselves as 'special' in negative way and believing they were different or abnormal to their peers without a chronic health condition.

> *"I don't know, probably because they are all normal and I am not." Colm (male, aged 12 years) (Benson et al., 2015)*

Although two of these studies were of lower methodological quality (Lewis & Parsons, 2008; Benson, Lambert, Gallagher, Shahwan, & Austin, 2015), their findings are consistent with papers of higher quality.

Of the eight studies, some CYP acknowledged that differences between peers were normal. This included two female adolescents, several 9-10-year olds with epilepsy and female CYP's with cleft lip and cleft palate (MacLeod, 2009; Moffat, et al., 2009; Tiemens, et al., 2013).

> *"everybody's messed up somehow one way or another" Katie (female) (MacLeod, 2009).*

Similarly, one quantitative study found that on average CYP reported neutral feelings towards their epilepsy on a self-stigma questionnaire (Benson et al., 2016).

Anticipated Stigma

Eight studies reported that CYP anticipated that stigmatising experiences would occur because of their chronic health condition (Benson et al., 2015; Dyson et al., 2010; Kirk & Hinton, 2019; Macleod, 2009; Moffat et al., 2009; Olsson et al., 2009; Tiemens et al., 2013, Wo et al., 2018). These included fears of their condition affecting friendships and relationships, being treated differently, being singled out by peers, being disliked, being subject to negative comments, being negatively labelled, being pitied or having general negative experiences because of their condition.

Perceived Stigma

Eight studies reported CYP perceived that others held stigmatising beliefs or opinions about them because of their chronic health condition (Benson et al., 2015; Dyson et al., 2010; Kirk & Hinton, 2019; Moffat et al., 2009; Olsson et al., 2009; Rhee et al., 2007; Tiemens et al., 2013; Wo et al., 2018). For example, peers or adults thinking they were different, unlikable, ugly or physically and mentally less competent than other CYP without a chronic health condition.

> *'I think that is also why I was shy because I didn't know what people were going to think because when you look like you are beautiful, people always talk to you more.'' (Tiemens et al., 2013).*

> *'When you say MS and when, the first thing that people think, disabled, in a wheelchair, about to die and old. Well, that's not true …. (Louise) (Kirk & Hinton, 2019)*

However, in one study, CYP reported both examples of perceived stigma and examples where they no longer became bothered by how others thought of them.

> *'Sometimes I felt kind of embarrassed (about my difference) but as I got older it has changed a lot because right now I don't really care what people think about me in terms of having a facial difference.'' (Tiemens, et al, 2013)*

Across the 14 papers, CYP of a range of ages and different chronic health conditions reported examples of self-stigma. In four studies CYP with epilepsy, orofacial clefts and celiac disease reported all three types of self-stigma (Benson et al., 2015; Moffat et al., 2009; Olsson et al., 2009; Tiemens et al., 2013).

3.3.2 Enacted Stigma

CYP reported experiences of enacted stigma because of their chronic health condition in 13 out of the 14 studies (Benson et al., 2015; Benson et al., 2016; Dyson et al., 2010; Kirk & Hinton, 2019; Kruger et al., 2014; Macleod, 2009; Masnari et al., 2012; Moffat et al., 2009; Olsson et al., 2009; Rhee et al., 2007; Tiemens et al., 2013; Usitalo, 2002; Wo et al., 2018). In 10 studies, CYP described examples of explicit enacted stigma such as bullying, physical violence, teasing, ridiculing and staring behaviours from both peers and adults related to their chronic health condition (Benson et al, 2015; Benson et al., 2016; Kirk & Hinton, 2019; Krüger et al., 2014; MacLeod, 2009; Moffat et al., 2009; Olsson et al., 2009; Rhee et al., 2007; Tiemens, et al., 2013; Wo et al., 2018)

> *"I used to get bullied, girls thought that I was some creature or alien from another planet, they used to say that to me and they used to say that I am not normal, the way I walk, the way I do things…" (Durriyah; Kirk & Hinton, 2019).*

Nine studies reported that CYP experienced implicit enacted stigma such as being socially excluded, having symptoms of their condition minimised or amplified by others, spreading of misinformation by peers, specific needs forgotten or ignored, and purposely being treated in a different way to others without chronic health conditions (Benson et al, 2015; Benson et al., 2016; Dyson et al., 2010; Kirk & Hinton, 2019; Krüger et al., 2014; MacLeod, 2009; Olsson et al., 2009; Rhee et al., 2007; Usitalo, 2002; Wo et al., 2018)

> *“…like some people don't really like to sit beside me …in case I would get a seizure or something like that.” Marie (female, aged 13 years; Benson et al., 2015)*

Findings indicated that CYP of different ages and with different chronic health conditions reported enacted stigma.

3.3.3 Concealment

Nine studies reported that CYP explicitly reported they wanted to conceal their condition from others (Benson et al., 2015; Benson et al., 2016; Dyson et al., 2010, Kruger et al., 2014; Macleod, 2009; Moffat et al., 2009; Olsson et al., 2009; Tiemens et al., 2013; Wo et al., 2018). This included CYP with concealable and less concealable conditions.

Methods of Concealing

Four studies commented on methods CYP used to conceal their condition (Kruger et al., 2014; Macleod, 2009; Olsson et al., 2009; Rhee et al., 2007). This included minimising or downplaying the severity of the condition, hiding or covering features of the condition, avoiding situations in which the condition would become apparent to others, attempting to pass as normal or avoiding conversations in which their condition would be discussed. There were two studies which suggested that some CYP did not conceal their condition from others. Kruger et al., (2014), found that 75.6% of CYP with vitiligo made very few or no attempts to conceal their condition to others, with Kirk and Hinton (2019), reporting that older CYP appeared to become more open about their MS as they adjusted to their illness.

Rationale for concealing

In six studies CYP explicitly discussed their rationale for concealing their chronic health condition (Benson et al., 2015; Kirk & Hinton, 2019; Lewis & Parsons, 2008, Macleod, 2009; Moffat et al., 2009; Olsson et al., 2009). In one study, concealment was reported under a main theme of ‘desiring normalcy’ and some CYP reported concealment helped them feel less different to others and avoided being treated differently or having restrictions placed upon them (Benson et al., 2015).

> *“Yes and I try to keep it private…Because it sort of makes me sad… I would feel upset; I would feel all the bad things” (Benson et al., 2015)*

In five studies, some CYP also reported that they concealed their chronic health condition to avoid feeling embarrassed about it (Benson et al., 2015; Lewis & Parsons, 2008; Moffat et al., 2009; Olsson et al., 2009; Tiemens, et al., 2013). CYP also reported they concealed their chronic health condition due to the expected responses of others. In four studies, some CYP reported that concealing their condition meant they did not need to manage the responses of others which could include others feeling distressed (Kirk & Hinton, 2019; Macleod, 2009; Olsson et al., 2009) and in two studies some CYP reported that concealing avoided having to have conversations about their condition (Kirk & Hinton, 2019; Olsson et al., 2009). In two studies, some CYP reported they concealed their chronic health condition in case those they disclosed to then told others (Lewis & Parsons, 2008; Moffat et al., 2009).

Overall, CYP appeared to conceal their condition to avoid negative feelings in themselves or to avoid the responses of others. There were no differences in concealment rationale across CYP of different ages or chronic health conditions.

3.3.4 Disclosure

Selective Disclosure

In seven studies, there was a theme of selective disclosure, in which CYP disclosed to close friends or peers, teachers or others outside of the nuclear family (Benson et al., 2015; Dyson et al., 2010; Kirk & Hinton, 2019; Lewis & Parsons, 2008; Macleod, 2009; Moffat et al., 2009; Wo et al., 2018). Of these seven studies, 22 CYP with epilepsy reported they had told their close friends (Moffat et al., 2009) and 90% of CYP with SCD had told at least one person they viewed as significant at school, such as a peer or teacher (Dyson et al., 2010).

Forced Disclosure

A common theme in nine of the studies was forced disclosure due to the presence of symptoms in front of others or noticeable treatment regimens (Benson et al., 2015; Dyson et al., 2010; Kirk & Hinton, 2019; Lewis & Parsons, 2008; Macleod, 2009; Moffat et al., 2009; Olsson et al., 2009; Rhee et al., 2007; Wo et al., 2018).

> *"What really gets you is if they see you having a fit" (Lewis & Parsons, 2008)*

Of these eight studies, five studies of generally high quality reported a similar theme; that CYP who reported that an unplanned disclosure had occurred, also reported increased feelings of self-stigma, including feelings of embarrassment and difference (Lewis & Parsons, 2008; Macleod 2009; Moffat et al., 2009; Olsson et al., 2009; Rhee et al., 2007). Furthermore, in four of the eight studies, CYP described experiencing enacted stigma following an unplanned disclosure, for example being teased, avoided or bullied (Benson et al., 2015; Kirk & Hinton, 2019; Macleod, 2009; Woo et al., 2018)

> *“Like, I used to take steroids, like, a lot of them, they used to say, oh, watch out, Nadia's going to come and punch you with her mega muscles” (Nadia: Kirk & Hinton, 2019)*

Rationale for Disclosure

In six studies, there were explicit examples of why CYP chose to disclose their chronic health condition (Benson et al., 2015; Dyson et al., 2010; Kirk & Hinton, 2019; Lewis & Parsons, 2008; Macleod, 2009; Moffat et al., 2009).

Two studies reported that CYP who disclosed did so for practical support (Benson et al., 2015; Kirk & Hinton, 2019), CYP in one study who disclosed, disclosed for emotional or peer support (Wo et al., 2018) and CYP in another study reported they disclosed for both practical and emotional or peer support (Lewis & Parsons, 2008; Macleod, 2009). Moffat et al. (2009), specifically reported that two CYP reported they disclosed their epilepsy to ensure their safety.

Advantages to Disclosing

Eight studies reported that some CYP discussed the advantages of disclosing their condition to others (Dyson et al., 2010; Kirk & Hinton, 2019; Lewis & Parsons, 2008; Macleod, 2009; Moffat et al., 2009; Olsson et al., 2009; Rhee et al., 2007; Wo et al., 2018). Of these studies, many CYP reported that peers became more supportive or helpful (Kirk & Hinton, 2019; Wo et al., 2018). Dyson et al. (2010) specifically reported that three CYP found peers to be more helpful following a disclosure. Similarly, Macleod (2009), reported all CYP in the study gained peer support from disclosing their chronic health conditions. Moffat et al. (2009), reported that four CYP described friends had been more supportive and seven CYP described teachers had been more supportive. Rhee et al., (2009), reported that some CYP said their disclosure positively changed the behaviour of others with two CYP reporting specifically:

> *"In the locker room, we'll use perfumes all the time, like, and they spray it and they don't care, and you're like, "dude, I have asthma, can you please stop?"(1) "My friends don't smoke around me because I lecture them every time they do.' (2) (Rhee, et al., 2007)*

Disadvantages to Disclosing

Five studies reported disadvantages of disclosing chronic health conditions (Dyson et al., 2010; Kirk & Hinton, 2019; Macleod, 2009; Olsson et al., 2009; Rhee et al., 2007). Of these four studies, Dyson et al., (2010) reported that nineteen CYP reported bad experiences when disclosing to peers and seventeen reported bad experiences when disclosing to teachers. Rhee et al., (2009) reported that some CYP reported their symptoms were minimised by teachers. In addition, Macleod (2009), found that two CYP had concerns that their disclosure to a teacher had been shared with other teachers and peers without their permission. Olsson et al. (2009), reported a theme emerged from their interviews with CYP of being the centre of attention in an unwelcome way due to having to disclose their dietary requirements. CYP in this study reported feeling frustrated even when concerns of others were well-meaning.

> *'Fanny: "Then they freak out and believe that you can't eat anything..." (Olsson, et al., 2009).*

A common theme which emerged from four of the five studies was that when CYP disclosed their condition, they received responses of pity or distress from others which triggered feelings of guilt, frustration or distress in the CYP themselves (Dyson et al., 2010; Kirk & Hinton, 2019; Macleod, 2009; Rhee et al., 2007)

> *"My guy friend who's like my brother, he came in and he just burst into tears…And it was heart-breaking because I felt mad for making them upset" (Louise) (Kirk & Hinton, 2019)*

Overall, the findings suggest that CYP generally make decisions to disclose their condition based on necessity or following an unplanned revealing of their condition, and that there can be positive and negative outcomes to disclosing. There were no differences in disclosure reports across CYP of different ages or chronic health conditions.

3.3.5 Relationship between Stigma and Concealment & Stigma and Disclosure

Two quantitative studies with robust methodology directly measured the relationship between stigma and concealment. They reported that greater perceived, internalised (Benson et al., 2016) and enacted stigma (Kruger et al., 2014) were significantly positively correlated with greater concealment strategies. Five high-quality qualitative studies reported that some CYP engaged in concealment strategies as a way of avoiding both self-stigma and enacted stigma (Benson et al., 2015; Kirk & Hinton, 2019; Macleod, 2009; Rhee et al., 2007; Wo et al., 2018).

No studies explicitly looked at the relationship between self or enacted stigma and disclosure, however, as previously reported in earlier findings, there are examples where some CYP in eight studies reported increased feelings of self-stigma and greater enacted stigma following a forced disclosure (Benson et al., 2015; Kirk & Hinton, 2019; Lewis & Parsons, 2008; Macleod 2009; Moffat et al., 2009; Olsson et al., 2009; Rhee et al., 2007; Woo et al., 2018).

3.3.6 Concealability

An unexpected theme emerged from the experiences of CYP in relation to their condition. In 11 studies CYP described the extent to which their condition could be concealable to others due to its physical characteristics (Benson et al., 2015; Benson et al., 2016; Kirk & Hinton, 2019; Lewis & Parsons, 2008; Macleod, 2009; Masnari et al., 2012; Olsson et al., 2009; Rhee et al., 2007; Tiemens et al., 2013; Usitalo, 2002; Wo et al., 2018). Seven studies reported that some CYP reported they could conceal their condition due to the invisibility of symptoms (Benson et al., 2015; Dyson et al., 2010; Kirk & Hinton, 2019; Kruger et al., 2014; Lewis & Parsons, 2008; Olsson et al., 2009; Wo et al., 2018). Two of these studies reported that the ability to conceal visible cues of their condition avoided feelings of perceived or anticipated stigma (Dyson et al., 2010; Kruger et al., 2014). Whereas in four studies, CYP reported that the invisibility of a condition could be a barrier to disclosure (Benson et al., 2015; Kirk & Hinton, 2019; Lewis & Parsons, 2008; Olsson et al., 2009; Rhee et al., 2007) because it could trigger misconceptions of the illness or that CYP might not be believed when they raised concerns or made disclosures amongst peers or teachers. Of these four studies, two described specific themes CYP reported, that the invisibility of epilepsy maintained the silence and lack of awareness around the condition (Benson et al., 2015; Lewis & Parsons, 2008).

Out of the 14 studies included in this review, four of these recruited samples of CYP with less concealable conditions, for example conditions such as vitiligo (Kruger et al., 2014), congenital and acquired facial differences (Masnari et al., 2012), orofacial clefts (Tiemens et al., 2013) and cranio-facial conditions (Usitalo 2002). Findings from one of these studies reported that 83% of CYP experienced enacted stigma due to visible manifestations of their condition and that there was lower satisfaction in physical appearance which was significantly associated with higher enacted stigma experiences (Usitalo, 2002). Another study found that CYP with larger facial differences were significantly more likely to experience enacted stigma and that having a facial difference covering over 25% of the face was a predictor for greater enacted stigma experiences (Masnari et al., 2012).

Findings suggest that concealability of a condition could impact concealment and disclosure processes amongst CYP who live with different types of chronic health conditions, including those which are more or less concealable.

4. Discussion

4.1 Study Aims

This systematic review had three key aims: 1) Describe the forms of stigma experienced by CYP with chronic health conditions, 2) Discuss any methods, rationale or implications CYP report for both concealing a chronic health condition and disclosing a chronic health condition, 3) Explore whether there is a possible connection between stigma experiences by CYP and concealment and/or disclosure of chronic health conditions. From the author's knowledge it is the first review to bring together experiences of CYP with different chronic health conditions and consider experiences of stigma, concealment and disclosure directly related to living with a chronic health condition. Unlike other reviews or empirical papers, it distinctly describes different types of stigma that CYP experience, separates the constructs of concealment and disclosure which are usually described as one entity, and describes possible relationships between stigma, concealment and disclosure. An unexpected theme to emerge was that of concealability and that the ability to conceal or hide a chronic health condition may impact CYP's experiences of stigma, concealment and disclosure.

4.2 1) Forms of Stigma Experienced

In the studies identified, CYP with chronic health conditions reported experiences of self-stigma and enacted stigma. Internalised stigma was reported by CYP in nine studies and both anticipated stigma and perceived stigma was reported by CYP in eight studies. There were no patterns to suggest that certain chronic health conditions or CYP of certain ages reported different self-stigma experiences. Enacted stigma appeared to be the most prevalent form of stigma and was reported in 13 out of 14 studies. This is in line with previous research suggesting that CYP often encounter adverse experiences with others because of their chronic health condition (McMurray et al., 2001; Nahal et al., 2019). It may also reflect that CYP find it easier to discuss concrete examples that elicit feelings of stigma, such as bullying or teasing, rather than internalised feelings of difference which are likely to have more subtle triggers, for example, the absence of chronic health conditions in the media.

The findings lend themselves to the wider literature on living with a stigmatised identity (Earnshaw & Chaudoir, 2009) and suggest CYP with different chronic health conditions at different ages and stages of development appear to have similar experiences of stigma. Experiencing stigma and living with a chronic health condition are risk factors for psychological distress (Quinn & Chaudoir, 2009) and co-morbid mental health difficulties (Gamwell et al., 2018; Pinquart & Shen, 2011). This could suggest that CYP with a chronic health condition who have stigmatising experiences are at a greater risk of experiencing psychological difficulties.

4.3 2) Concealment and Disclosure Processes

The desire to conceal their chronic health condition from others was a common theme reported by CYP in most studies. Methods of concealment included minimising or masking symptoms and avoiding particular situations or conversations about their condition. In some studies, CYP reported that they concealed their condition to feel more normal and less embarrassed, to prevent differential treatment and to avoid evoking distress or unwelcome interest from others. Concealing a chronic health condition has been found to adversely impact peer relationships and physical safety of CYP (Velsor-Friedrich et al., 2004). These findings can help to understand the rationale CYP choose to conceal and the common themes amongst several different chronic health conditions.

Although many CYP reported they had disclosed their chronic health condition to people outside of their family, they appeared to have selectively disclosed out of necessity or safety to gain practical or emotional support. In addition, eight studies described instances where CYP were forced to disclose their condition due to the onset or noticeable management of symptoms, which could lead to experiences of enacted stigma and self-stigma. Advantages of disclosing included others becoming more supportive and helpful. Previous research found similar themes and that CYP who disclosed their chronic health conditions noticed better peer relationships (Kaushansky et al., 2017; McMurray et al., 2001). However, CYP reported that disclosing their chronic health condition could also trigger unwelcome attention, distress and pitying responses from others. The responses CYP receive following a disclosure is likely to influence their future disclosure decisions, particularly if they receive a negative response. Therefore, it is important to understand the types of negative responses CYP experience and consider how to prevent or manage their impact. No patterns emerged which suggested that particular chronic health conditions differ in their concealment and disclosure processes.

4.4 3) Relationship between Stigma, Concealment and Disclosure

Two quantitative studies found direct relationships between greater self-stigma (Benson et al., 2016) and greater enacted stigma (Krüger et al., 2014) and more concealment in CYP with chronic health conditions. In six qualitative studies, a common theme CYP expressed was that of concealing their condition to avoid experiences of self-stigma and enacted stigma. There were no studies which explicitly reported direct relationships between stigma and disclosure, but a theme across eight of the studies was that CYP reported greater self or enacted stigma experiences, particularly following a forced disclosure.

Similar findings have been observed in studies of adults living with chronic health conditions, with reported relationships between stigma and concealment (Lee et al., 2017) and stigma and disclosure (Kaushansky et al., 2017). This review suggests there is likely to be a relationship between stigma and concealment, and stigma and disclosure in CYP with chronic health conditions.

4.5 Concealability

In eleven studies, CYP referred to how concealable their chronic health condition was to others. Several CYP with conditions such as epilepsy, MS, vitiligo, sickle disorder and celiac disease, commented that they could conceal their condition, or that invisibility of symptoms allowed them to conceal it from others (Benson et al., 2015; Dyson et al., 2010; Kirk & Hinton, 2019; Kruger et al., 2014; Lewis & Parsons, 2008; Olsson et al., 2009; Wo et al., 2018) and that this could be useful in avoiding anticipated stigma (Dyson et al., 2010; Kruger et al., 2014). However, it was also reported that the invisibility of symptoms could be a barrier to disclosure and facilitate silence around chronic health conditions. From this it could be tentatively hypothesised that having a less visible chronic health condition may reduce stigma experiences in the short-term but contribute to maintain and reinforce stigmatising experiences longer-term. Two studies which looked at experiences of CYP with facial differences (Masnari et al., 2012) and cranio-facial conditions (Usitalo 2002), found that enacted stigma experiences were specifically related to the visible characteristics of their conditions.

In this review it was not possible to compare concealable and less concealable conditions directly. However, the findings suggest there is complex relationship between concealability and stigma, which echoes research in adult samples (Elstad et al., 2010; Kaushansky et al., 2017).

4.6 Contextualising the research

The majority of studies included in this review are from western countries using western approaches to health and medicine which are typically attuned to acute illnesses and treating such conditions effectively (Quintner et al., 2008; Thorne, 2008). Therefore, when chronic conditions present differently to acute conditions this may not be in line with how illnesses are perceived in the wider society and trigger unhelpful opinions and beliefs. For example, two studies in this review described times CYP with Sickle Cell Disease and epilepsy were forced to reassure others their condition was not contagious (Dyson et al., 2010; Wo, et al., 2018). These misconceptions are likely to impact the way that CYP with chronic health conditions are viewed in communities.

In the last decade there has been a rise in digital usage and social media (Ofcom., 2019). There is evidence that social media can influence body image perceptions and enhance negative social comparisons (Fardouly et al., 2015; Marengo et al., 2018). Many of the studies included in this review were published at least five to ten years ago when social media was not so prevalent in the daily experiences of CYP. It is possible that if similar studies were replicated now, reported stigma, concealment and disclosure practices might be influenced by other experiences.

4.7 Limitations of this Review

Firstly, the review only included English language studies which may have narrowed the studies selected and limit the generalisation of the findings to non-English populations.

Secondly, six studies were included in the review which focused on CYP with epilepsy. No other chronic health conditions were represented more than once. This does reflect the current field of literature whereby there is a focus on experiences of CYP with epilepsy. Epilepsy has been the target of discrimination and prejudicial behaviour across cultures throughout history (De Boer et al., 2008) and this could explain why there is a greater prevalence of studies exploring stigma experiences of CYP with epilepsy. It is possible that the findings and themes of this review may have been influenced by the experiences of CYP with epilepsy. Although it was not possible to make comparisons due to the mix of study designs, there were no notable patterns to suggest that CYP with other chronic health conditions had different experiences of self-stigma, enacted stigma, concealment, disclosure or concealability.

Finally, although multiple databases were searched, it is possible relevant studies were missed. Qualitative studies in particular can be more complex to identify as search terms often do not correspond well to titles and abstracts (David, 2002). Stigma can also be conceptualised in many ways and few of the studies included referred to explicit definitions of stigma during data collection, measurement or analysis. Although this review has attempted to synthesise stigma experiences based on explicit definitions, it is possible that other researchers may have used alternative definitions and synthesised this information differently.

4.8 Implications

4.8.1 Research Implications

This review indicates that there are common experiences of self-stigma, enacted stigma, concealment and disclosure among CYP with different chronic health conditions. However, the literature is in its infancy and because of this, it is difficult to make comparisons and extrapolate whether differences between chronic health conditions do exist. For example, the theme of concealability emerged from this review, which could suggest that CYP with more or less concealable conditions could have different experiences of stigma, concealment or disclosure. Therefore, it is important for more research, with larger sample sizes and a range of study designs to explore stigma, concealment and disclosure in CYP with different chronic health conditions.

Evidence suggests that stigma is not an individual process and intersects with community and cultural values (Remedios & Snyder, 2015; Stangl et al., 2019; Turan et al., 2019). This suggests that experiences of stigma in CYP may vary in different communities. Future research could explore the impact of stigma, and the role of concealment and disclosure in different cultures and communities. This could help to better understand the experiences of CYP and take account of intersectionality and other factors which may be similar or different in the experiences of CYP with chronic health conditions.

4.8.2 Clinical Implications

This review indicates that CYP with chronic health conditions as young as six experience enacted stigma from others and perceive, anticipate and internalise stigma themselves. It is important for clinicians working with CYP and their families to be aware of the different ways in which stigma presents. From extrapolating the findings in this review and considering psychological models generally, cognitive or behavioural strategies may be more effective with feelings of self-stigma whereas coping strategies or role-plays centred around help-seeking following bullying and teasing may be more relevant for enacted stigma experiences.

Findings from the review also indicated that unplanned or forced disclosures due to the onset of symptoms or treatment may enhance feelings of self-stigma and increase the likelihood of enacted stigma from others. Within generic and specialist paediatric settings, it could be important to provide opportunities for CYP and their families to prepare for such situations and mitigate the impact of difficult feelings if and when the unplanned disclosure occurs. Additionally, some CYP reported instances where both concealing their chronic health condition and disclosing their condition led to a misunderstanding of symptoms that could threaten their physical safety (Dyson et al., 2010; Rhee et al., 2007). When a chronic health condition is diagnosed or during follow up appointments, it would be useful for clinicians to have regular conversations with families to discuss effective ways to develop relationships with schools or other systems which allows for a better understanding of the CYP’s chronic health condition and its impact. This could mitigate against some of the difficult experiences reported by CYP, particularly with teachers, and help to ensure that individual needs are understood, can be responded to safely and appropriately and reduce the burden and stigma attributed to CYP.

Conclusions

This review has found that CYP experience both self and enacted forms of stigma directly related to living with a chronic health condition. Enacted stigma, the negative judgement and treatment by others, was most prevalent. The review has identified that concealment and disclosure are not mutually exclusive concepts and that although many CYP desire to keep their condition concealed from others they also participate in disclosure strategies at the same time. The unexpected finding of concealability has indicated that there could be differences in how CYP experience stigma, concealment and disclosure depending on how concealable their condition is to others. More research is needed to explore this area and to directly measure the relationships between stigma, disclosure and concealment.

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Part 2: Empirical Paper

The Experiences of Children and Adolescents with Physical Health Conditions: The impact of Self-Stigma and Visible Difference on Concealment, Illness Attitudes and Psychosocial Difficulties.

Abstract

Aims & Background: There is an increased prevalence of psychological distress and poorer long-term outcomes for Children and Young People (CYP) living with physical health conditions. To better understand the psychological processes of living with a physical health condition, this study aimed to explore the impact of stigma and visible or less visible differences on concealment, illness attitudes and emotional and peer difficulties in CYP with physical health conditions.

Method: 61 CYP were recruited from a specialist national hospital and completed three validated questionnaires to investigate self-reported self-stigma, concealment beliefs and illness attitudes. Measures of visibility, psychosocial difficulties and relevant demographic and medical information were obtained from caregivers.

Results: CYP with visible physical health conditions reported significantly poorer illness attitudes and more emotional and peer difficulties with large and moderate effect sizes respectively. Higher reported self-stigma significantly predicted greater concealment, poorer illness attitudes and more emotional and peer difficulties in CYP.

Conclusions: The findings suggest that condition-specific factors such as visibility can affect the attitudes CYP have towards their physical health condition, their emotional wellbeing and peer relationships. The visibility of a condition and stigma perceptions should be taken into account when supporting CYP with physical health conditions and their families.

1. Introduction

1.1 Background

Children and young people (CYP) with long term or recurring physical health conditions are at greater risk for developing psychological difficulties (Gamwell et al., 2018; Holmbeck et al., 2002; Hysing et al., 2007). Many psychological difficulties in childhood and adolescence persist into adulthood (Kessler et al., 2005). Therefore, identifying problems early and providing effective support to CYP is essential. Developing psychological interventions for CYP with physical health conditions to improve their psychological wellbeing requires greater understanding of the specific factors which influence the relationship between CYP and their condition.

There is some evidence that suggests that how CYP relate to their physical health conditions can influence their illness-related behaviours and subsequently their psychosocial functioning in childhood and adulthood. Two constructs that have been used to understand how CYP relate to their condition are stigma and illness attitudes. CYP report frequent stigma experiences (Austin et al., 2004; Strauss et al., 2007; Taylor et al., 2008), and have both positive and negative illness-related attitudes (Funderburk et al., 2007). Additionally, some findings indicate that condition specific factors such as the visibility of a physical health condition and illness-related behaviour such as concealment (Benson et al., 2015) may be related to stigma experiences (Kaushansky et al., 2017; Pachankis et al., 2018). However, research in this area is limited, with qualitative findings and epilepsy-specific studies dominating the field.

The aim of this study is to enhance the understanding of CYP with physical health conditions by exploring the presence of and relationships between stigma, concealment, illness related beliefs and psychosocial difficulties. This study also aims to progress the existing literature by investigating CYP with physical health conditions other than epilepsy, and to explore the impact of visibility of physical health conditions on the psychological factors above.

1.2 Stigma

Stigma can be defined as an undesired difference (Goffman, 2009) and individuals are usually stigmatized when they possess a difference which is not viewed as normal in society (Stangor & Crandall, 2000). Experiencing any form of stigma is associated with a range of adverse outcomes and can negatively affect psychological wellbeing (Quinn & Chaudoir, 2009) and social development of CYP (Mak et al., 2007; Walker & Reznik, 2014). However, stigma is a complex phenomenon and has been conceptualised in different ways that focus on different aspects of the stigma experience. Within the research there is a prevailing focus on the impact of enacted stigma, specifically the experiences of discrimination, prejudice or negative stereotyping from others (Earnshaw & Chaudoir, 2009; Kinsler et al., 2007).

The specific focus of this study will be on self-stigma, which refers to the internalisation of external stigmatizing messages (Quinn & Earnshaw, 2013; Teh et al., 2014), the perception that others hold stigmatizing beliefs about them (Van Brakel, 2006) and anticipation of stigmatizing experiences (Earnshaw et al., 2013; Quinn & Chaudoir, 2009). There is evidence that CYP with physical health conditions experience self-stigma (Kirk & Hinton, 2019;

Olsson et al., 2009; Wo et al., 2018) and that it can contribute to psychological distress in CYP (Austin et al., 2004; Lambert & Keogh, 2015) and mental health difficulties in adulthood (Quinn & Earnshaw, 2013). However, the literature has heavily focused on the experiences of CYP with epilepsy (Austin et al., 2004; Benson et al., 2015b), and often reports general experiences of stigma, which can include both enacted and self-stigma experiences. Understanding more about the impact of self-stigma could enable services to develop more effective ways of reducing these negative feelings and associated adverse outcomes in CYP.

1.3 Visibility

Many physical health conditions cause visible changes. During childhood and adolescence there is an increased focus on appearance and body image (Crerand et al., 2017; McCabe et al., 2005), and having a visible difference compared to peers can present psychological, social and developmental challenges for CYP (Rumsey & Harcourt, 2007). Several studies have already considered the impact of visible conditions on enacted stigma such as bullying and teasing (Ablett & Thompson, 2016; Feragen & Stock, 2016). There is evidence that visibility of a condition may also impact self-stigma. Some CYP report greater self-stigma because their condition is visible to others (Masnari et al., 2012; Strauss et al., 2007). By comparison, other research with young adults with visible differences suggest they report less shame, the emotion elicited when one experiences socially rejecting experiences such as stigma, and embarrassment compared to those who have less visible conditions (Kaushansky et al., 2017). Similarly, there is evidence to suggest that self-stigma is higher when conditions are less visible (Dos Santos et al., 2017) and that CYP anxiously anticipate negative responses from others if the condition suddenly becomes visible, for example, a seizure occurring in a CYP with epilepsy (Benson et al., 2016). The existing literature is unclear about the relationship between the visibility of a condition and stigma and no studies with CYP have compared reported self-stigma in CYP with visible or less visible physical health conditions.

1.4 Illness Attitudes

CYP with physical health conditions develop illness related beliefs and attitudes based on their experiences with physical symptoms, treatment regimens and the practical limitations imposed by their condition (Ramsey et al., 2016). In the literature illness attitudes is defined as a concept taking into account several cognitive processes, including how CYP appraise the uncertainty of their condition, its intrusiveness and the perceived control and self-confidence they believe they have in managing the condition (Austin & Huberty, 1993; Hoff et al., 2002; Ramsey et al., 2016). Studies exploring illness attitudes often use the terms positive or negative illness attitudes to describe the cognitive appraisals that CYP may demonstrate towards their condition. For example, CYP may be described as having more negative illness attitudes if they report feeling ‘bad’ because of their condition or report feeling like their condition is their fault (Austin & Huberty, 1993; Heimlich et al., 2000; Ramsey et al., 2016). Although creating labels of ‘negative’ and ‘positive’ may cause value-based judgements to be made of the attitudes and beliefs CYP hold towards their illness, there is some evidence to suggest differentiating different illness attitudes can be helpful in understanding other experiences of CYP with chronic health. For example, some research suggests that CYP with chronic health conditions who report more negative illness attitudes can have poorer outcomes managing their condition and report poorer quality of life (Edgar & Skinner, 2003; Heath et al., 2017; Law et al., 2014; Nabors et al., 2010). Studies have also found associations between self-stigma in CYP and illness attitudes. CYP with chronic health conditions reporting greater self-stigma also report more negative illness attitudes towards their epilepsy (Austin et al., 2004; Funderburk et al., 2007). Furthermore, some evidence indicates that particular features of a physical health condition can impact illness attitudes (Austin & Huberty, 1993; Ryan et al., 2012). For example, some CYP with more visible conditions report more limitations on everyday life because of their conditions, than CYP with less visible conditions (Rasmussen et al., 2018; Walker et al., 2004). However, other studies have also found that children with less visible conditions hold negative views towards their condition (Austin et al., 2004). Currently there is no research explicitly comparing illness attitudes in CYP with visible and less visible conditions.

1.5 Concealment

Concealment is a strategy that CYP with physical health problems are known to employ in an attempt to avoid negative attention from others (Bachmann et al., 2009; Jantzen et al., 2009). It is important to understand concealment beliefs, as several studies indicate that concealment of any identity, including health conditions, can impact psychological and physical quality of life (Quinn et al., 2017), peer relationships and management of health conditions (Rhee et al., 2007; Wo et al., 2018). There is evidence to suggest that feelings of self-stigma, because of a physical health condition, may influence how likely an individual is to conceal the condition from others (Kaushansky et al., 2017; Lee et al., 2017). Research has found that CYP with physical health conditions conceal their condition to avoid feeling embarrassed and being treated differently (Bachmann et al., 2009; Houston et al., 2000), which are typical examples of self-stigma. Another study found a specific relationship between self-stigma and concealment, with CYP who reported greater self-stigma more likely to conceal their condition from others (Benson et al., 2016). However, the sample did not use a validated measurement of concealment. If CYP conceal their condition due to feelings of self-stigma, this is likely to lead to an unhelpful cycle and reinforce unhelpful stigmatising beliefs. There could also be differences in concealment in those who have more or less visible physical health conditions. Young adults with visible conditions were reported to be more comfortable speaking about their physical health condition than those with less visible conditions (Kaushansky et al., 2017). Similarly, adults with less visible disabilities reported that they were more likely to conceal their condition than those with visible disabilities (Hernandez, 2011).

While some studies have considered the impact of visibility on concealment behaviours, there are no studies directly comparing concealment of physical health conditions in CYP with visible or less visible conditions. In addition, although some research has considered the association between stigma and concealment, in CYP, most samples are of CYP with epilepsy.

1.6 Emotional and Peer Difficulties

It is well established that CYP with physical health conditions are more likely to experience emotional distress and co-morbid mental health difficulties compared to peers without a physical health condition (Gamwell et al., 2018; Pinquart & Shen, 2010). Research with both adult and CYP studies suggest there is a relationship between psychological wellbeing and self-stigma (Baker et al., 2018; Quinn et al., 2014). Specifically, studies with CYP report greater feelings of self-stigma are associated with increased anxiety, depressive symptoms (Austin et al., 2004) and behavioural difficulties (Funderburk et al., 2007). There is also evidence to suggest that the visibility and appearance of physical health conditions may increase the risk of psychosocial difficulties in CYP including emotional, behavioural and interpersonal difficulties (Rasmussen et al., 2018; Rumsey & Harcourt, 2007). CYP with health conditions which affect their appearance have been reported to withdraw socially and show symptoms of depression and anxiety (Dennis et al., 2006). Similarly, CYP with less visible conditions have also reported emotional distress and difficulties with peer relationships (Austin et al., 2004). It is particularly relevant to understand both self-stigma and the visibility of conditions in relation to emotional and peer difficulties in CYP due to the long-term impact of distress and interpersonal difficulties on mental health and help-seeking (Gulliver et al., 2010; Kessler et al., 2005).

1.7 Summary

There is currently some preliminary research that suggests self-stigma is associated with illness attitudes, concealment and psychosocial difficulties in CYP with physical health conditions. However, the majority of studies have been qualitative, mainly using CYP with epilepsy and often report general experiences of stigma, rather than highlighting the impact of self-stigma explicitly. In addition, there are no current studies which have looked into the visibility of a physical health condition as a specific condition related factor. Due to the higher prevalence of psychological difficulties in CYP with physical health conditions and the risk of these difficulties progressing into mental health conditions (Kessler et al., 2005), greater understanding of the experiences of CYP is crucial. Therefore, this study aimed to understand the influence of 1) the visibility of a physical health condition and, 2) self-stigma related to a physical health condition on the psychological processes of living with a physical health condition in childhood and adolescence.

Research Questions

1) Are there significant differences in self-stigma, concealment, illness attitudes and emotional and peer difficulties between CYP with visible and less visible physical conditions?

2) Are there significant associations between stigma and concealment, illness attitudes, and emotional and peer difficulties, and does the visibility of a condition impact the associations?

2. Method

2.1 Design

The study was a quasi-experimental cross-sectional design which collected questionnaire data from CYP with physical health conditions and their caregivers. CYP completed three validated self-report questionnaires asking about self-stigma, concealment and illness attitudes. Caregivers completed one validated parent-report questionnaire about emotional and peer difficulties observed in their child, and a study specific questionnaire concerning demographic and medical questions including a specific question asking how visible their child's physical health condition was to others.

The study used a convenience sample and tested the following hypotheses: 1) Are there significant differences in self-stigma, concealment, illness attitudes and emotional and peer difficulties between CYP with visible and less visible physical conditions 2) Is stigma a significant predictor of concealment, illness attitudes and emotional and peer difficulties, and does the visibility of a condition influence this relationship. Tests of difference were used to analyse differences between visible and less visible physical health conditions and child reported self-stigma, concealment and illness attitudes, and parent reported emotional and peer difficulties. Bivariate correlations and hierarchal regressions were used to analyse relationships between self-stigma, visibility and concealment, illness attitudes and emotional and peer difficulties.

2.2 Joint Thesis Declaration

This was a joint project working in collaboration with Claire Hackford, who was exploring stigma and psychosocial difficulties of CYP with physical health conditions, specifically the associations between CYP and caregivers (Hackford, 2020; Appendix D).

2.3 Sample

Participants were 61 CYP and their parents who attended appointments in a specialist paediatric hospital between October 2019 and May 2020 for dermatological and urological conditions. CYP were identified by hospital records and included if they fulfilled the following criteria: 1) Aged between 8-14, 2) attended outpatient appointments at Great Ormond Street Hospital, 3) had an ongoing dermatological or urological condition requiring treatment or follow-up appointments, 4) attended the appointment with their caregiver. CYP were excluded if: 1) they were acutely unwell, 2) had a developmental delay or learning disability which meant they could not complete questionnaires independently, 4) CYP or caregiver did not have sufficient English reading or writing skills needed to complete the survey. 242 CYP who fit age and disorder criteria were identified from hospital data, 123 caregivers gave initial consent and a total of 61 CYP and caregivers completed data collection, resulting in a response rate of 25.2%. More detailed information on the data collection and attrition process is presented in Figure 1. The reasons caregivers gave for not participating are reported in Table 1.

The final sample comprised of 61 CYP and their caregivers. Overall the sample was made up of 31 female (50.9%) and 30 male (49.2), 35 (57.4%) aged between 8.08 – 14.92 years with an average age of 12 years (S.D. = 1.77). 26 of CYP were reported to have visible conditions and 35 were reported to have less visible conditions. Visibility was defined based on the following question '*if someone walked past your child in the street would they notice they had a physical health problem*' answered by the caregiver. More detailed information on measures used is discussed in section *'2.4 Measures'.* Full demographic and clinical information for caregivers and CYP is presented in Table 2 and Table 3.

Figure 1:

Data Collection Process

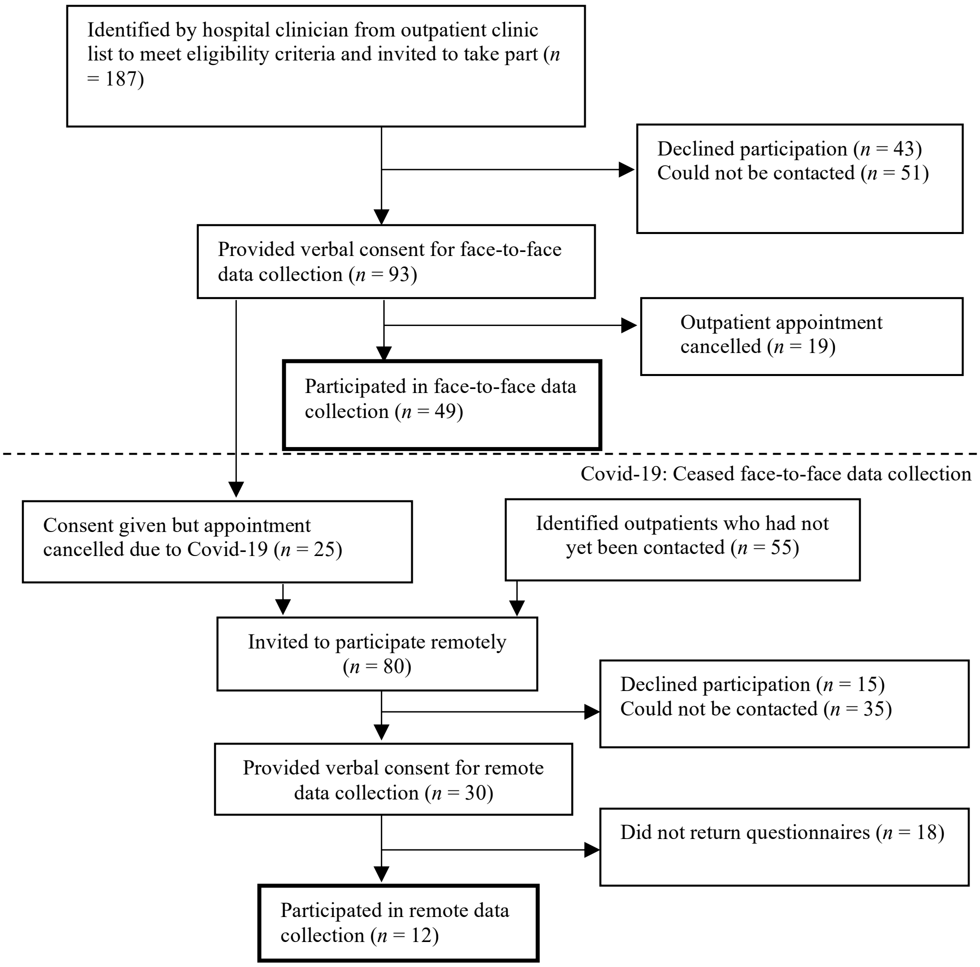

Table 1:

Declined Participation

Caregiver reported rationale	
No reason given by caregiver	16
Caregiver specifically reported that CYP did not want to take part	11
Caregiver concerned questionnaire would have a negative effect on CYP	7
CYP had co-morbid condition making it difficult to take part	7
Declined remote data collection	8
Caregiver perceived study was not relevant to CYP	5
Too busy	4

**n = 58*

Table 2:

Demographic Profile of Caregivers

Demographics	
Caregiver Gender	
Female	51 (83.6%)
Male	10 (16.4%)
Caregiver Ethnicity	
White British or White other	46 (75.5%)
Mixed	1 (1.6%)
Asian/Asian British	8 (13.1%)
Black/Black British	4 (6.6%)
Other	1 (1.6%)
PNS	1 (1.6%)

Table 3:

Demographic and Medical Profile of CYP

Demographic Variables	Visible PHC[a] (n= 26)	n	Less Visible PHC[a] (n = 35)	n
Gender				
Female	61.5%	16	42.9%	15
Male	38.5%	10	57.1%	20
Age				
Mean	12.39	26	11.67	35
SD	1.92		1.60	
Ethnicity				
White British or White other	73.1%	19	77.1%	27
Asian/Asian British	15.4%	4	8.6%	3
Black/Black British	7.7%	2	5.7%	2
Mixed			5.7%	2
Other	3.8%	1		
PNS[b]			2.9	1
Age of onset				
Present at Birth	38.5%	10	40%	14
Younger than 5 years	42.3%	11	34.3%	12
5 years +	19.2%	5	25.7%	9

Hospital Attendance				
1-2 times per year	34.6%	9	60%	21
3-10 times per year	50%	13	34.3%	12
10 + times per year	15.4%	4	5.7%	2
Type of clinic				
Dermatology	88.5%	24	31.4%	11
Urology	11.5%	3	68.6%	23
Type of PHC[a]				
Eczema	57.7%	15	11.4%	4
Epidermolysis Bullosa (EB)	15.4%	4	14.3%	5
Psoriasis	15.4%	4	5.7%	2
Bladder Condition	7.7%	2	28.6%	10
Kidney Condition	3.8%	1	40%	14

PHC[a] – physical health conditions, PNS[b] – prefer not to say,

2.4 Measures

2.4.1 Patient and Public Involvement & Engagement (PPI-E)

In order to ensure the measures chosen for this study were appropriate for CYP, a PPI-E phase was conducted in collaboration with the CYP service user consultation committee of the specialist paediatric hospital where the data was collected. Seven children aged between 8-14 years-old agreed to complete the questionnaire pack independently and they were asked 1) If the questionnaires were understandable, 2) If there were any other barriers to completion. Overall the feedback was positive with some minor changes to terminology requested. As these changes did not affect the standardisation of the questionnaire, they were made prior to submission to research governance application.

2.4.2 Child Report Measures

Child Stigma Scale (CSS) (Appendix E): An eight-item questionnaire (Austin et al., 2004) was used to measure self-stigma in CYP. CYP were asked to rate items such as, '*How often do you feel people may not want to be friends with you if they know you have x condition?*' using a 5-point Likert scale from 1 ('Never') to 5 ('Very Often). The total score is calculated by summing the items and dividing them by the total number of five items. Total scores range between 1-5, with higher scores reflecting greater perceptions of self-stigma. Initially validated on 9-14 years-old with epilepsy by the developers of the scale (Austin et al., 2004), this questionnaire was selected as it has also been adapted for use across other physical health conditions (Gamwell et al., 2018) and children with mental health difficulties (Kaushik et al., 2017). The developers of the scale reported good internal consistency, α = 0.81 and construct validity (Austin et al., 2004).

The Secrecy Scale (Appendix F)*:* A seven-item scale (Kaushik et al., 2017) was used to measure concealment in CYP. CYP were asked to rate such items as '*When I meet people for the first time, I make a special effort to keep the fact that I have a health condition to myself'* using a 4-point Likert scale ranging from 1 ('I disagree a lot') to 4 ('I agree a lot)*'.* The total score is calculated by summing the items (including reverse coded items) and dividing them by the total number of seven items, with higher scores reflecting greater concealment beliefs. The scale was taken from the *Paediatric Self-Stigmatisation Scale (PeadS) (*Kaushik et al., 2017) and for the purpose of this study, the questionnaire was adapted for use with children with physical health conditions by replacing the term *'difficult feelings and behaviour'* with *'medical condition'*. The questionnaire was initially validated on children aged 8-12 years-old with mental health conditions (Kaushik et al., 2017). This measure was chosen due to the accessible language for CYP and was reported to have good internal consistency in this population, α = 0.79 (Kaushik et al., 2017).

Child Attitude Towards Illness Scale (CATIS) (Appendix G): A thirteen-item questionnaire (Austin & Huberty, 1993) was used to measure illness attitudes. CYP were asked their positive and negative attitudes about having a physical health condition. CYP are asked to rate items such as '*How bad or good do you feel it is to have x physical health condition'* using a 5-point Likert scale ranging from 1 (e.g. 'Very Good'*)* to 5 *(*e.g. 'Very Bad'). The total score is calculated by summing the items (including reverse coded items) and dividing by the total number of 13 items. The total score ranges from 1-5, where higher scores represent more positive illness attitudes. The questionnaire was selected due to its initial validation by the developers of the scale on children aged 8-17 years-old with a range of physical health problems (Heimlich et al., 2000). Previous research suggests the CATIS also has good internal consistency (α = .89) and test-retest reliability (Heimlich et al., 2000).

2.4.3 Parent Reported Measures

Demographic and Medical Profile (Appendix H): Caregivers were asked to complete a study specific questionnaire which included demographic questions about their child's age, gender, ethnicity and additional medical information about their child's physical health condition.

Emotional and Peer Difficulties (Appendix I): A ten-item scale (Goodman, 1997) was used to ask caregivers to rate their child on emotional and peer related items such as *'Many fears, easily scared' or 'Has at least one good friend'* using a 3-point Likert scale 1 *('not true'*) to 3 ('*certainly true'*). Items relating to emotional symptoms and peer relationships are summed and calculated to form the emotional and peer difficulties subscale (Goodman, 1997), with scores ranging from 0-20 with higher scores indicating greater emotional and peer difficulties. The scale was part of the Strengths and Difficulties Questionnaire (SDQ) and evidence suggests the emotional and peer difficulties scale is appropriate for use in epidemiological research (Goodman et al., 2010). The developers of the questionnaire reported good internal consistency on emotional symptoms scale (α = .80) but lower internal consistency for the peer relationship (α = .40) sub-scale (Goodman, 2001). This scale was selected as it is widely used in paediatric settings (Flannery et al., 2018; Pitchforth et al., 2019) and emotional distress and peer difficulties are particularly relevant to CYP with physical health conditions (Gamwell et al., 2018).

Visibility (Appendix H)*:* Because of the complexity in defining visible difference in an objective way, research has used a functional definition of visible difference (Soon, 2015). The most common index used is whether someone walking down the street would notice a difference or disfigurement on another individual. As part of the demographic questions, all caregivers were asked '*If someone walked past your child in the street would they notice they had a physical health problem*" (Appendix H). Children whose caregiver answered 'yes' were allocated to the visible group and children whose caregivers answered 'no' were allocated to the less visible group.

2.4.4 Reliability of Questionnaire Measures

Reliability analyses were performed to assess the psychometric properties of the validated questionnaires when administered to this current sample of CYP and their caregivers (n=61). The CSS (self-stigma) (α = .89), the Secrecy Scale (concealment) (α = .86) and the CATIS (illness attitudes) (α = .87) all had good internal consistency, while the Emotional and Peer Difficulties scale had acceptable internal consistency (α = .75), as determined by Cronbach's alpha.

2.5 Ethical considerations

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

2.6 Power Analysis

Previous studies found significant differences ($p<0.05$) between CYP with visible and non-visible physical health conditions and experiences of shame (Mason, 2010) and significant differences ($p<0.05$) across CYP with different physical health conditions and concealment of their conditions (Westbrook et al., 1991) with large effect sizes (Cohen's $d = 0.79$ and 0.73 respectively). Data from shame related research was deemed acceptable to use as shame and stigma are related social constructs and found to be associated in the perception of health-related beliefs (Rose et al., 2017). Considering the effect sizes and the planned statistical tests (e.g. tests of difference), several power analyses were carried out using G* Power 3.1 software (Faul et al., 2007), specifying alpha at 5% and desired power at 80%. The sample size required to compare CYP with visible and non-visible differences was between 27 and 31 CYP per group, 54 and 62, in total, respectively.

Studies of CYP who had physical health conditions also reported significant relationships ($p<0.05$) between self-stigma and greater concealment of physical health conditions (Benson et al., 2016), self-stigma and poorer illness-related attitudes (Austin et al., 2004), and self-stigma and greater emotional difficulties (Austin et al., 2004), with large effect sizes (r = 0.57, -0.60, 0.48 respectively). Considering the effect sizes and the planned statistical tests (e.g. correlations and multiple regressions), several power analyses were carried out using G* Power 3.1 software (Faul et al., 2007), specifying alpha at 5% and desired power at 80%. The analyses revealed that sample size needed was between 17-25 CYP.

To satisfy the research questions of the study and the planned statistical analysis, a total sample size of 62 CYP was required, 31 with visible and 31 with less visible differences.

2.7 Procedure

CYP who fit disorder and age criteria were identified by a member of the care team from electronic dermatology and urology outpatient clinic lists approximately four weeks prior to their scheduled outpatient appointment. An information pack with details of the study was sent to the patient's recorded home address that contained a Letter of Invitation to Participate (Appendix L), a Participant Information Sheet for caregivers (Appendix M), and a Participant Information Sheet for CYP (Appendix N). A telephone call was made by the researchers approximately two weeks prior to the CYP's next outpatient clinic appointment in the specialist paediatric hospital. In this call, the researchers re-checked that CYP fit the disorder and age criteria, discussed participation with the caregiver and gained verbal consent. CYP and their caregivers were met by a researcher on the day of their outpatient clinic appointment in the clinic waiting room. The researcher discussed the questionnaires and ensured the CYP and caregiver could give informed assent and informed consent. Written consent from the caregiver (Appendix O) and written assent from the CYP (Appendix P) was collected. Caregivers and CYP completed the questionnaires independently in a quiet area of the outpatient waiting room. The researcher was available to answer any questions and provide support during and after completion of the questionnaires. Caregivers and CYP received a debrief where they were signposted to relevant services if appropriate. Caregivers were also made aware of how to contact the principal researcher if they had any future questions and those who provided consent were sent information about the outcomes of the study.

Due to the CO-VID 19 pandemic, all face-to-face non-urgent clinic outpatient appointments in the hospital were cancelled during March, April and May 2020 and data during this period was collected remotely. The same information sheets were sent approximately four weeks prior to the cancelled appointment or the new telephone clinic appointment. Caregivers were contacted on the telephone as before to discuss remote participation and gain verbal consent. Caregivers were asked if they would prefer to participate over the telephone or via email. If conducted over the telephone the researcher went through the consent form and questionnaire with the caregiver and CYP, if completed via email, consent forms and questionnaires were sent to the caregivers, completed independently and returned to the researcher. If conducted over the telephone, the same support and debrief information was provided as face to face data collection. If completed via email, the researchers followed up with a telephone call to provide the debrief. Data protection and data governance policies issued during the pandemic by the specialist paediatric hospital involved in the study were followed. All participants (caregivers and CYP) were issued with a participation code which was recorded on their surveys. All questionnaire data was uploaded using the code to SPSS version 26 with relevant protective software installed.

2.8 Statistical Analyses

All data collected was cleaned and checked for errors prior to statistical analysis. Normality tests were performed on the four variables (self-stigma, concealment, illness attitudes and emotional and peer difficulties) to satisfy the assumptions of the planned statistical tests. Kolmogorov-Smirnov statistic indicated that scores on self-stigma, concealment, illness attitudes were normally distributed (Appendix Q). Emotional and peer difficulties scores were not normally distributed ($p<0.01$) with skewness of 0.95 (SE = 0.31) and kurtosis of 0.62 (SE = 0.61, Therefore on tests of difference, non-parametric tests were used for this variable. To manage the impact of extreme values, z-scores were computed for all four key variables (self-stigma, concealment, illness attitudes and emotional and peer difficulties) and none were found to be > 3.29 suggesting there were no outliers present (Appendix R).

T-tests and ANOVAs or the non-parametric equivalent were performed on all demographic information to test for any significant differences between groups. Post-Hoc testing with Bonferroni corrections were employed to find between group differences. Performing these tests allowed for significant demographic factors to be controlled for in other analyses.

T-tests were used to assess group differences between CYP with visible and less visible physical health conditions and reported self-stigma, concealment and illness-related attitudes. Non-parametric Mann-Whitney U tests were used to explore differences between CYP with visible and less visible physical health conditions and reported self-stigma and emotional and peer difficulties. ANCOVAs were used to control for the impact of possible co-variates when significant differences were observed, due to the categorical nature of the variables, some of the test assumptions were violated. These are discussed in the results

Bi-variate correlational analysis was used to explore the relationships between self-stigma, concealment, illness attitudes and emotional and peer difficulties. Hierarchal multiple regression models were used to analyse the relationships between self-stigma, concealment, illness attitudes and emotional and peer difficulties, to explore the influence of visibility on these relationships and to account for the impact of co-variates.

3. Results

3.1 Sample Characteristics

The mean scores CYP reported for self-stigma, concealment, illness attitudes and emotional and peer difficulties are presented in Table 4. On the Child Stigma Scale, the mean response reported was between ‘sometimes’ or ‘often’ (M = 2.40, SD = 0.94) indicating that CYP felt stigmatised because of their physical health condition. On the Secrecy Scale, the mean response reported was between disagree and agree (M = 2.42, SD = 0.68), indicating that CYP held neutral beliefs about concealing their condition from others. On the Illness Attitudes scale, the mean response (M = 3.22, SD = 0.71) indicated that CYP held neutral to positive illness attitudes. The Emotional and Peer Difficulties scale reported a mean score of 6.30 (SD = 0.77), with higher scores indicating greater emotional and peer difficulties.

Table 4:

Self-stigma, Concealment, Illness attitudes and Emotional & Peer difficulties (Mean/SD) of the current sample compared to published samples

Variable	Self-Stigma (Child Stigma Scale)		Concealment (Secrecy Scale)		Illness Attitudes (CATIS)		Emotional Difficulties (SDQ subscale)		Peer Difficulties (SDQ subscale)	
Score Range	*(0-5)*		*(0-4)*		*(0-5)*		*(0-20)*			
	M	SD	M	SD	M	SD	M	SD	M	SD
Current Sample *n =61*	2.40	.94	2.42**	.68	3.22	.71	4*	2.6	2.29*	2.25
Austin et al. (2004) *n =170*	2.24	.88	-	-	-	-	-	-	-	-
Kaushik et al. (2017) *n = 156*	-	-	2.70**	.69	-	-	-	-	-	-
Austin et al. (2006) *n =170*	-	-	-	-	3.34	.65	-	-	-	-
de la Cruz et al. (2018) *n = 8434*	-	-	-	-	-	-	4.7*	2.8	3.6*	2.4

***P<0.01, * p<0.05*

One sample t-tests were used to compare the current sample to other clinical samples. There were significant differences between the average concealment score of CYP in this sample compared to a sample of CYP with which the measure was developed t (60) = -3.16, p<0.01, with the current sample reporting lower concealment scores on average. There was also a significant difference between emotional and peer difficulties compared to a large CYP UK paediatric sample, t (60) = -2.10, p < 0.04, t (60) = -4.54, p <0.01, (de la Cruz et al., 2018), suggesting that CYP in the current study had fewer emotional and peer difficulties.

3.1.1 Demographic and Medical Variables

T-tests, ANOVA's, Mann Whitney U and Kruskal-Wallis tests were used to explore the influence of demographic and medical variables on, self-stigma, concealment, illness attitudes and emotional and peer difficulties. This allowed for demographic and medical variables to be entered as co-variates in later analysis and control for the impact of confounding variables. Face-to-face data collection and remote data collection were also compared. All findings are presented in Appendix S and significant results reported.

Concealment scores significantly differed between CYP with eczema (n=19), Epidermolysis Bullosa (EB) (n=9), psoriasis (n=6), bladder condition (n=12) and kidney condition (n=15), F (3,57) = 1.66, p <0.05. Post hoc comparisons found higher concealment in CYP with psoriasis (M = 2.98, SD = 0.51) compared to CYP with kidney conditions (M = 2.10 = SD = 0.58), mean difference of 0.88, 95% CI [0.01 – 1.75], p <0.05.

Illness attitudes were significantly poorer in females (M = 3.02, SD = 0.67) than males (M = 3.43, SD = 0.70), M = - 0.41, 95% CI [-0.76 – (-0.05)], t (59) = -2.29, p < 0.05. Illness attitudes were significantly poorer in CYP attending dermatology clinics (M = 2.95, SD = 0.58) than CYP attending urology clinics

(M = 3.54, SD = 0.73), M = - 0.55, 95% CI [-0.93 – (-0.25)], t (59) = 3.50, p < 0.01. Illness attitudes significantly differed between CYP with eczema (n=19), EB (n=9), psoriasis (n=6), bladder conditions (n=12) and kidney conditions (n=15), F (3,57) = 3.18, p <0.05. Post hoc comparisons found poorer illness attitudes in CYP with eczema (M = 3.00, SD = 0.60) compared to CYP with bladder conditions (M = 3.73, SD = 0.64), mean difference of -0.73, 95% of CI [-1.41 – (-0.05), p<0.05, and poorer illness attitudes in CYP with psoriasis (M = 2.72, SD = 0.82) compared to CYP with bladder conditions (M = 3.73, SD = 0.64), mean difference -1.01 [-1.94 – (-0.87)], p<0.05.

Emotional and peer difficulties signifcantly differed between CYP who attended hospital 1-2 times per year (n = 30), 3-10 times per year (n=25) and more than 10 times per year (n=6), X^2 (2) = 8.49, p < 0.05. Pairwise comparisons using Bonferroni corrections found that CYP who attended hospital 3-10 times a year had statistically significantly more emotional and peer difficulties (Mdn = 7.00) than CYP who attended hospital 1-2 times a year (Mdn =. 4.00), P<0.05.

3.2 Research Question 1:

Are there differences in self-stigma, concealment, Illness Attitudes, emotional and peer difficulties in CYP with visible and less visible physical health conditions?

Self-stigma, concealment and illness attitudes were normally distributed and independent t-tests were performed to detect any differences between CYP with visible and less visible physical health differences (Table 5). The emotional and peer difficulties scale was not normally distributed, and a Mann Whitney U test was performed to detect any differences in CYP with visible or less visible physical health conditions (Table 6) and emotional and peer difficulties.

Table 5:

T-test: Differences in Self-Stigma, Concealment and Illness Attitudes based on Visibility

	Visible (n=26)		Less Visible (n=35)		t (59)	p	Cohen's d
	M	SD	M	SD			
Self-Stigma	2.61	.82	2.23	1.0	1.60	0.12	0.42
Concealment	2.37	.70	2.47	.68	-.55	0.59	0.15
Illness Attitudes	2.96	.63	3.41	.72	-2.55	0.02*	0.67

*n = 61, *p<0.05*

CYP with visible physical health conditions reported poorer illness attitudes (M = 2.96. SD = 0.63) compared to CYP with less visible conditions (M = 3.41, SD = 0.72), a statistically significant difference, M = -0.45, 95% CI [-0.80 – (-0.10)], t (59) = - 2.55, p <0.05, with a large effect size (Cohen, 1988). There were no significant differences observed on the measures of self-stigma (p =0.12) and concealment (p=0.59).

Table 6:

Mann Whitney U: Differences in Emotional & Peer Difficulties based on Visibility

Independent Variable	Visible	Less Visible	U	z	p	r
	Mdn	Mdn				
Emotional and Peer Difficulties	7.5	4	245.5	-3.071	.00**	-.39

*n = 61, **p<0.01*

CYP with visible differences (Mdn = 7.5) had signficantly greater emotional and peer difficulties than CYP with less visible differences (Mdn = 4), U = 245.5, z = -3.07, p <0.01 with a medium effect size (Cohen, 1988).

3.2.1 Impact of co-variates

Further analyses were performed on the significant differences observed between CYP with visible and less visible differences. Separate one-way ANCOVA's were performed and demographic variables assessed in the preliminary analysis that were found to differ significantly on the measures of illness attitudes and emotional and peer difficulties were entered as co-variates. Due to the sample size only one co-variate was selected to ensure that the analysis could be adequately powered (Field, 2009).

Illness attitudes were found to be significantly different across three demographic variables, gender, clinic type (dermatology or urology) and physical health conditions (eczema, EB, psoriasis, bladder conditions & kidney conditions) To satisfy the assumptions of the ANCOVA, chi-square tests of association were performed to ensure that the variables of gender, clinic type and physical health condition were not significantly associated with visibility (Table 7).

Table 7:

Chi square tests: Association of Gender, Clinic type and PHC[a] with Visibility

	Chi square (X)	P value	Effect size (Cramer's V)
Gender	2.08	.15	.19
Clinic Type	19.66	.00**	.57
PHC[a]	22.92	.00**	.62

***p <0.01, PHC[a] – physical health conditions*

There was a statistically significant association between visibility and clinic type, $X^2(1) = 19.66$, $p<0.01$; and between visibility and physical health condition, $X^2(4) = 22.92$, $p<0.01$ with large effect sizes (Cohen, 1988). There was no significant association between visibility and gender, $X^2(1) = 2.08$, $p=0.15$. Therefore, in line with the assumptions, only gender was entered into the ANCOVA as a co-variate. Due to the nominal characteristics of the variable gender, several assumptions of the ANCOVA were hard to test such as checking for a linear relationship between the co-variate (gender) and the dependent variable (illness attitudes) and checking for homoscedasticity. All other assumptions were satisfied. An ANCOVA was performed and after adjusting for gender, a statistically significant difference was still observed in illness attitude between children with visible and less visible conditions, $F(1, 58) = 4.86$, $p = 0.03$, partial $\eta^2 = 0.08$, indicating that CYP with visible physical health conditions had significantly poorer illness attitudes than those with less visible physical health conditions.

Emotional and peer difficulties were found to be significantly different across the demographic variable hospital attendance. To satisfy the assumptions of the ANCOVA, chi-square tests of association were performed to ensure that hospital attendance was not significantly associated with visibility (Table 8).

Table 8:

Chi square tests: Association of Hospital Attendance with Visibility

	Chi square (X)	P value	Effect size (Cramer's V)
Hospital Attendance	4.27	0.12	0.27

There was no significant association between visibility and hospital attendance $X^2(2) = 4.27$, $p=0.12$, and hospital attendance was entered into the ANCOVA as a co-variate. Due to the nominal characteristics of the variable hospital attendance, several assumptions of the ANCOVA were harder to test such as checking for a linear relationship between the co-variate (hospital attendance) and the dependent variable (emotional and peer difficulties) and checking for homoscedasticity. All other assumptions were satisfied. An ANCOVA was performed, after adjusting for hospital attendance, a statistically significant difference remained in emotional and peer difficulties between children with visible and less visible conditions, F (1, 58) = 5.63, $p < 0.05$, partial $\eta^2 = 0.09$, indicating that CYP with visible conditions had significantly greater emotional and peer difficulties than those with less visible conditions.

3.3 Research Question 2

Is there an association between self-stigma and concealment, illness attitudes and emotional and peer difficulties, and does the visibility of a condition influence this relationship?

3.3.1 Bivariate Correlations to determine the relationships between variables

Before looking at more complex analysis, bivariate correlations were carried out to examine the relationships between the variables and to ensure none of the test variables were highly correlated. Variables were entered within one matrix and are reported in Table 9.

Table 9:

Pearson Correlations of Self-Stigma, Concealment, Illness Attitudes, Emotional and Peer Difficulties

Variable	*n*	*M*	*SD*	1	2	3	4
1. Self-Stigma	61	2.40	.94	-	.61**	-.69**	.46**
2. Concealment	61	2.42	.68	.61**	-	-.31*	.28*
3. Illness Attitudes	61	3.42	.71	-.69**	-.31*	-	-.40**
4. Emotional/ Peer Difficulties	61	6.30	4.04	.46**	.28*	-.40**	-

***p<0.01 *p<0.05*

Self-stigma is positively correlated with concealment (r =.61, p < 0.01), emotional and peer difficulties (r=.46, p<0.01) and negatively correlated with illness attitudes (r =.69, p < 0.01). None of the correlation coefficient's were above the threshold (r ≥ .80) deemed to suggest collinearity between variables (Field, 2009).

3.3.2 Hierarchal Regression Analyses

A series of hierarchal multiple regression analyses were run to determine if there was an association between self-stigma and concealment, illness attitudes and emotional and peer difficulties. Visibility (visible or less visible physical health condition) was added as an independent variable in all analyses to assess if explained additional variance in each regression model. Co-variates identified in the preliminary analysis were added to the relevant regression model to account for the effect of confounding variables. Due to the sample size, no more than four independent variables were added to the regression models (Field, 2009).

3.3.2 Self-stigma and Concealment

Two hierarchical multiple regressions were run to determine if the addition of visibility (visible or less visible) explained additional variance in the association between self-stigma and concealment, above self-stigma alone and to control for the effect of physical health condition (eczema, EB, psoriasis, bladder conditions & Kidney conditions) (Table 10).

Table 10:

Hierarchical Multiple Regression Predicting Concealment from Self-Stigma, Visibility & PHC

	Concealment			
	Model 1		Model 2	
Variable	B	ß	B	ß
Constant	1.37		.86	
Self-stigma	.44**	.61	.47**	.65
Visibility			.277	.20
R^2	.37		.41	
F	34.33**		19.91	
(Δ) R^2	.37		.04	
(Δ) F	34.33		3.84	

	Concealment					
	Model 1		Model 2		Model 3	
Variable	B	ß	B	ß	B	ß
Constant	2.45		1.23		0.86	
PHC	-.01	-.024	.04	.09	-.02	-.04
Self-stigma			.45**	.62	.47**	.65
Visibility					.31	.23
R^2	.00		.38		.41	
F	.03		17.40**		13.10	
(Δ) R^2	.00		0.37		.03	
(Δ) F	.03		34.75		3.19	

*** $p<0.01$, PHC[a] – physical health conditions*

Self-stigma was an independent predictor of concealment even after controlling for the impact of CYP's physical health condition.

The full model of self-stigma and visibility to predict concealment (Model 2) was statistically significant, $R^2 = 0.41$, F (2,58) = 19.91, $p < 0.01$, adjusted $R^2 = 0.39$. The addition of visibility accounted for an additional 4% of variance but was not a statistically significant difference, $R^2 = 0.04$, F (1,58) = 3.84, p= 0.06. The full model of self-stigma and visibility to predict concealment (Model 3) when controlling for type of physical health condition was also statistically significant, $R^2 = 0.41$, F (3,57) = 13.10, p <0.01, adjusted $R^2 = 0.38$.

Overall the models indicate that greater self-stigma scores is associated with greater concealment scores and when type of physical health condition and visibility is kept constant, for every increase of 1 on the concealment score, mean self-stigma scores would also increase by 0.47.

3.3.3 Self-Stigma and Illness Attitudes

Two hierarchical multiple regressions were run to determine if the addition of visibility (visible or less visible) explained more of the variance in the association between self-stigma and illness attitude above self-stigma alone and to control for the effects of gender and type of clinic (dermatology or urology) (Table 11).

Table 11:

Hierarchical Multiple Regression Predicting Illness Attitudes from Self-Stigma, Visibility & Type of Clinic

Illness Attitudes				
	Model 1		Model 2	
Variable	B	ß	B	ß
Constant	4.46		3.98	
Self-stigma	-.52**	-.69	-.49**	-.65
Visibility			0.26	.18
R^2	.47		.50	
F	52.28**		29.23	
(Δ) R^2	.47		.03	
(Δ) F	52.28		3.74	

Illness Attitudes						
	Model 1		Model 2		Model 3	
Variable	B	ß	B	ß	B	ß
Constant	3.14		4.29		4.11	
Gender	.25	.17	.14	.10	.14	.10

Type of Clinic	-.51**	-.36	-.32*	-.23	-.27	-.19
Self-stigma			-.46**	-.61	-.46**	-.60
Visibility					.10	.07
R^2	.20		.54		.54	
F	7.21**		22.17**		16.53	
(Δ) R^2	.17		.51		.51	
(Δ) F	7.21		41.90		.36	

*** $p<0.01$, *$p<0.05$*

Self-stigma was an independent predictor of illness attitudes even after controlling for gender and type of clinic. The full model of self-stigma and visibility to predict illness attitudes (Model 2) was statistically significant, R^2 = 0.50, F (2,58) = 29.23, p <0.01, adjusted R^2 = 0.46. The addition of visibility explained an additional 3% of the variance but this was not a significantly statistical difference, R^2 = 0.03, F (1,58) = 3.74, p= 0.06. The full model of self-stigma and visibility to predict illness attitudes (Model 3), when controlling for gender and type of clinic, was statistically significant, R^2 = 0.54, F (4,56) = 16.28, p <0.01, adjusted R^2 = 0.51. Overall, Model 3 indicates greater self-stigma scores are associated with poorer illness attitudes and that when gender, type of clinic and visibility is kept constant, for every decrease of 1 on the illness attitudes score, mean self-stigma scores would increase by 0.46.

3.3.4 Self-Stigma and Emotional and Peer Difficulties

Two hierarchical multiple regressions were run to determine if the addition of visibility explained more of the variance in the association between self-stigma and emotional and peer difficulties above self-stigma alone and to control for the effect of hospital attendance (1-2 times, 3-10 times & 10 + times per year) (Table 12).

Table 12:

Hierarchical Multiple Regression Predicting Emotional and Peer Difficulties from Self-Stigma, Visibility & Hospital Attendance

	Emotional and Peer Difficulties			
	Model 1		Model 2	
Variable	B	ß	B	ß
Constant	1.56		5.80	
Self-stigma	1.98**	.46	1.73**	.40
Visibility			-2.31*	-.29
R^2	.21		.29	
F	15.72**		11.76**	
(Δ) R^2	.21		.08	
(Δ) F	15.72		6.38	

	Emotional and Peer Difficulties					
	Model 1		Model 2		Model 3	
Variable	B	ß	B	ß	B	ß
Constant	2.48		-1.18		2.48	
Hosp. Attendance	2.38**	.39	2.02**	.33	1.70*	.28
Self-stigma			1.77**	.41	1.61**	.37
Visibility					-1.76	-.22
R^2	.15		.32		.35	
F	10.67**		13.54**		10.73	
(Δ) R^2	.15		.17		.04	
(Δ) F	10.67		14.05		3.81	

*** p<0.01, *p<0.05*

Self-stigma was an independent predictor of Emotional and Peer Difficulties even when controlling for the impact of hospital attendance.

The full model of self-stigma and visibility to predict emotional and peer difficulties (Model 2), was statistically significant, $R^2 = 0.29$, F (2,58) = 11.76, p <0.01, adjusted $R^2 = 0.26$. The addition of visibility accounted for an additional 8% of variance which was a statistically significant difference, $R^2 = 0.08$, F (1,56) = 6.38 p <0.05. The full model of self-stigma and visibility to predict emotional and peer difficulties (Model 3), when controlling for hospital attendance, was also statistically significant, $R^2 = 0.35$, $F(3,57) = 10.73$, $p < 0.01$, adjusted $R^2 = 0.33$. Overall, Model 3 indicates greater self-stigma scores are associated with greater emotional and peer difficulties and that when hospital attendance and visibility is kept constant, for every increase of 1 on the emotional and peer difficulties scale, mean self-stigma scores would also increase by 1.61.

4. Discussion

4.1 Summary

The first aim of this study was to explore if CYP with visible health conditions reported differences in self-stigma, concealment, illness attitudes and emotional and peer difficulties psychological than CYP with less visible conditions. The study found that CYP with visible physical health conditions had significantly poorer illness attitudes and emotional and peer difficulties than CYP with less visible physical health conditions. There were no significant differences between CYP with visible and less visible physical health conditions and reported self-stigma or concealment. The second aim of this study was to explore the relationships between self-stigma, concealment, illness attitudes and emotional and peer difficulties in CYP with physical health conditions. Self-stigma was found to be a significant predictor of concealment beliefs, illness attitudes and emotional and peer difficulties. Visibility only influenced the relationship between self-stigma and emotional and peer difficulties.

4.2 Sample characteristics

CYP in this sample reported similar perceptions of self-stigma and illness attitudes compared to previous samples of CYP with physical health conditions such as epilepsy (Austin et al., 2006; Austin et al., 2004). This indicates that findings from epilepsy studies, of which there are many, could be applicable to CYP with other physical health conditions. CYP in this study reported significantly lower concealment beliefs compared to the sample on which the questionnaire was validated (Kaushik et al., 2017). However, the original questionnaire specifically assessed concealment of mental health conditions and was not specific to concealment beliefs in physical health conditions. Similarly scores on the emotional and peer subscales were significantly better in this study than in other paediatric samples (de la Cruz et al., 2018). The comparison sample consisted of data collected in routine clinical practice across a range of mental health and physical health conditions. It is possible that differences have been found due to the self-selected nature of this current sample and the sole focus on physical health conditions. Therefore, caution should be taken when applying these findings to CYP with more severe emotional and peer difficulties.

4.3 Medical Factors

During the preliminary analysis, there were several medical variables that significantly differentiated the sample on some of the psychological variables. CYP with psoriasis had higher concealment scores than CYP with kidney difficulties and poorer illness attitudes than CYP with bladder difficulties. Similarly, CYP with eczema had poorer illness attitudes than CYP with bladder difficulties. Group sizes were small; however, it could be tentatively suggested that concealment beliefs and illness attitudes may be different in certain conditions, a finding observed in other studies (Ryan et al., 2012). CYP who attended dermatology clinics were found to have poorer illness attitudes than CYP who attended urology clinics. Lastly, CYP who attended hospital 3-10 times per year were found to have more emotional and peer difficulties than CYP who attended hospital once or twice a year. More frequent hospital attendance can be indicative of more complex medical conditions and this finding could indicate that CYP with more complex physical health conditions have more emotional and peer difficulties.

4.3 Differences in CYP with Visible and Less Visible Physical Health Conditions

The results showed that CYP with visible conditions reported poorer illness attitudes and more emotional and peer difficulties than children with less visible conditions. To the authors knowledge this is the first study to observe these findings. This suggests that CYP who have a visible physical health condition are more likely to hold more negative attitudes towards their condition than CYP with less visible physical conditions. This finding is supported by previous studies which found CYP with visible physical health conditions reported negative illness attitudes (Rasmussen et al., 2018; Walker et al., 2004). Possessing negative attitudes towards a physical health condition can have wider implications. For example, negative illness attitudes have been associated with poorer health management (Nabors et al., 2010) and quality of life in CYP (Heath et al., 2017), experiences which are themselves likely to perpetuate negative illness attitudes. However, earlier analysis also indicates there is likely to be an overlap between visibility and the type of clinic (dermatology or urology) CYP attend on difference in illness attitudes. Visibility and clinic type were found to significantly associated and both CYP with visible conditions and CYP from dermatology were found to have poorer illness attitudes compared to CYP with visible conditions and CYP from urology clinics respectively. This is not surprising as 88.5% of CYP with visible physical health conditions also attend dermatology clinics. It could be suggested that CYP with dermatological conditions are also likely to have more visible conditions which contribute to the influence on illness attitudes. But it is not possible to conclude that the visibility of a condition fully accounts for the differences in illness attitudes alone and it is likely that both visibility and type of clinic contribute to this difference.

The results also indicate that CYP with visible health conditions have more emotional and peer difficulties than CYP with less visible physical health conditions. This finding aligns with prior studies which found that the visibility of physical health conditions may increase the risk of psychosocial difficulties in CYP (Dennis et al., 2006; Rasmussen et al., 2018). Emotional and peer difficulties in childhood can be associated with behavioural problems, social isolation and poorer academic attainment (Fantuzzo et al., 2005; Rubin et al., 2009; Valiente et al., 2012). Furthermore, they can increase the risk for developing mental health conditions as an adult (Kessler et al., 2005). Therefore, while it is well established that having a physical health condition in childhood and adolescence increases the risk of emotional distress and impacts peer relationships (Gamwell et al., 2018; Pinquart & Shen, 2010), the finding of this study suggests that having a visible difference may increase these risks further. Unlike findings with illness attitudes, there were no initial differences observed between clinic type (dermatology or urology) and emotional and peer difficulties in CYP. Therefore, it can be more confidently suggested that the visibility of a physical health condition accounted for the observed difference in emotional and peer difficulties.

Reported self-stigma and concealment did not significantly differ between CYP with visible and less visible physical health conditions. Previous studies have alluded to differences in stigma (Benson et al., 2016; Masnari et al., 2012) and concealment (Kaushansky et al., 2017) in CYP with visible and less visible conditions, however this was not observed in the current sample. It is possible that CYP do not internalise a sense of difference based on the visibility of their condition and therefore feelings of self-stigma are similar across CYP with visible and less visible conditions. Similarly, concealment beliefs and behaviours held by CYP with physical health conditions can be complex and influenced in many ways (Benson et al., 2015a; Olsson et al., 2009; Wo et al., 2018). It is also possible that the measure used to define visibility in this study did not capture subtle differences which may exist in reported self-stigma or concealment in CYP with more or less visible conditions.

4.4 Self-Stigma as a predictor of concealment, Illness Attitudes and Emotional & Peer Difficulties

Self-stigma was found to be the strongest predictor for concealment, illness attitudes and emotional and peer difficulties, even when accounting for the impact of co-variates and the influence of visibility. Findings suggest that CYP who report greater self-stigma are more likely to want to conceal their condition from others, show more negative attitudes towards their physical health condition and have more difficulties with emotions and peer relationships. The findings are consistent with other research in CYP with physical health conditions, which suggests that self-stigma can be associated with a range of psychological and behavioural process (Bachmann et al., 2009; Benson et al., 2016). Concealing a physical health condition can be particularly problematic for CYP as it can be a barrier to gaining practical and social support (Rhee et al., 2007; Wo et al., 2018). Furthermore, negative illness attitudes can have adverse implications on the management of a condition (Nabors et al., 2010), and emotional and peer difficulties can impact longer term mental health outcomes (Kessler et al., 2005).

The visibility of a condition significantly predicted emotional and peer difficulties independently, suggesting that CYP with visible conditions have more emotional and peer difficulties. Visibility also accounted for significantly more of the variance between the relationship of self-stigma and emotional and peer difficulties than self-stigma alone. Visibility was not a significant predictor of illness attitudes or concealment.

Overall, these results add to existing literature exploring the impact of stigma in CYP with physical health conditions. It specifically highlights the impact of self-stigma; the process in which stigma experiences are internalised to the self and on which future predictions and assumptions are based. It also suggests that the visibility of a condition may be an important factor to consider particularly in relation to illness attitudes and emotional and peer difficulties.

However, the findings of the study are solely based on self-reported questionnaires and may not accurately reflect the lived experiences of CYP. Additionally, the concealment questionnaire was developed for CYP with mental health difficulties and the self-stigma and illness attitudes questionnaire were developed for CYP with epilepsy. While the measures had been used in research with CYP reporting different physical health conditions, it is possible that they did not capture experiences of the CYP in this sample relevant to these constructs.

4.5 Research Implications

This study highlights the importance of considering specific illness related characteristics such as the visibility of a condition. Future research could explore the impact of visibility in several ways. In this study, only caregivers were asked to answer whether their child's condition was visible or not to others. While caregivers are important advocates for their child's emotional and physical health needs (Jordan et al., 2018), it would be useful to ask both caregivers and CYP directly whether they view their physical health condition as visible or not. Additionally, many physical health conditions can change in presentation over time (Rumsey & Harcourt, 2007) or become more or less visible suddenly, for example, seizures in epilepsy (Benson et al., 2015). One way to capture the changeability and complexity of visibility could be to use a scale. Another way could be to use different research designs such as focus groups or interviews, to explore with CYP if and when they feel their condition is visible or less visible and what this means to them. This could help to better understand the impact of visibility on CYP and explore factors that may protect and mitigate against difficult feelings and beliefs about having a visible or less visible condition.

The questionnaires used in this study used a range of items to explore self-reported self-stigma, concealment, illness attitudes and parent reported emotional and peer difficulties. If the same questionnaires are used in future research, it could be useful to do more detailed analysis on whether certain themes or factors emerge from the questionnaires. For example, if there were specific items on the attitudes towards illness scale which differed across groups, such as gender, age or type of physical health condition.

This study focused on aspects of self-stigma, future research could also look at enacted stigma, i.e. times that CYP have actually experienced discrimination or exclusion because of their physical health conditions (Earnshaw & Chaudoir, 2009; Kinsler et al., 2007), and how this may be different between CYP with visible and less visible conditions, and if there are different relationships between enacted stigma, illness attitudes, concealment and emotional and peer difficulties. Better understanding of enacted stigma could also help to prompt more systemic awareness and macro level changes.

4.6 Clinical Implications

The findings suggest that clinicians should be aware that CYP with visible physical health conditions could be at risk of developing negative illness attitudes and more emotional and peer difficulties. To mitigate these risks, clinicians working in paediatric settings could consider implementing means to better monitor if distress occurs. For example, being curious about how CYP feel about their conditions, working jointly with families and mental health clinicians to allow these conversations to be held sensitively without pathologizing CYP and their families, and/or referring CYP and their families to specific psychological services within or external to paediatric settings when appropriate.

If CYP are seen in clinical or paediatric settings, there is some evidence to suggest psychological interventions could be useful to manage distress. A specific compassion-focused intervention for adults with skin conditions found improvements in self-compassion and negative affect (Sherman et al., 2019). Furthermore, a brief evidence-based CBT intervention improved distress associated with experiences of enacted stigma such as teasing and bullying in CYP with appearance differences (Maddern et al., 2006). These interventions could be adapted to explore the impact of visibility and to specifically explore self-stigma, which could improve the outcomes for CYP with both visible and less visible conditions.

Alongside more individualised interventions, it is also important to consider systemic factors and how CYP might internalise messages and judgements (Kinsler et al., 2007). It could be helpful for health care professionals to discuss wider issues that may perpetuate stigma and to share good practice amongst colleagues and in systems around CYP, such as primary care services and schools.

4.7 Limitations

There are several limitations of the current study. Firstly, 71% of the self-selected CYP and caregiver sample were White British. Physical health stigma has been found to intersect with other stigmatised identities, such as ethnicity (Remedios & Snyder, 2015; Stangl et al., 2019; Turan et al., 2019) and therefore caution should be taken when generalising these findings more broadly to CYP from other ethnic groups.

The visibility of a condition can be complex (Rumsey & Harcourt, 2007; Benson et al., 2015) and this study used a simple parent-reported measure of visibility which may not have captured the variability and changeability of how visible a physical health condition can be. There was also an association between visibility (visible and less visible) and clinic type (dermatology and urology) which restricted the specific conclusions that could be made about the differences in illness attitudes in CYP. Prior research indicates that condition specific factors can influence illness attitudes in CYP (Ryan et al., 2012), which has been observed in this study. However, more research is needed to tease apart the effects of visibility and type of condition to understand more about the experiences CYP with physical health conditions including self-reported illness attitudes.

Lastly, data collection was interrupted by the COVID-19 pandemic. Remote methods were employed to continue the recruitment of CYP and their caregivers and there were no significant differences in CYP or caregivers who participated before or during the pandemic. However, this affected the desired sample size as many participants who provided verbal consent before the pandemic, either did not consent to remote participation or did not return the questionnaires remotely. Additionally, the response rate of 25.2% was fairly low overall and the findings are likely to reflect a bias in those participated in the study.

Conclusion

CYP with visible physical health conditions are more likely to have more negative feelings towards their condition and more emotional and peer difficulties than CYP with less visible physical health conditions. This suggests that there is a need to increase awareness amongst clinical services and provide avenues for early psychological support for CYP with more visible health conditions. CYP who report greater self-stigma around their physical health condition are also more likely to conceal their condition from others, have more negative feelings towards their condition and have more emotional and peer difficulties. Due to the long-term adverse outcomes which can be associated with these psychological and behavioural processes, targeted interventions are needed to reduce self-stigma in CYP with physical health conditions.

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Part 3: Critical Appraisal

1. Introduction

The purpose of this critical appraisal is to evaluate and reflect on some of the key issues encountered during the completion of the research project. I will discuss how the issues have shaped my understanding and how they could be addressed in the future. The critical appraisal will be detailed in two sections. Firstly, I will discuss my reflections throughout the process of completing the systematic review and the empirical study, including my personal experiences which may have shaped my ideas and my understanding of CYP with physical health conditions. Secondly, I will specifically discuss the research methodology of the empirical study, critique the measures chosen and consider ways data collection could have been more inclusive of diverse samples of CYP.

2. Personal Reflections

2.1 Establishing and Developing the Project

Feeling stigmatised is extremely common in individuals with physical health conditions (Van Brakel, 2006; Weiss et al., 2006) and can have a negative impact on quality of life (Earnshaw et al., 2012). I have a personal interest in this area as members of my immediate family live with physical health conditions. Before the project, I was aware of the challenges people with physical health conditions face; including managing the misconceptions of others, navigating complex health care systems and changing their routines and valued interests because of their health. Additionally, from job roles prior to the doctorate course, I worked with several young adults and adolescents with physical health conditions. They frequently reported feeling different or inadequate compared to their peers and did not feel understood when they talked about their condition to others. This led them to avoid certain situations, stop engaging in certain hobbies and interests, and created an underlying fear about how their condition would impact the rest of their lives. Through developing my understanding of the literature, these young adults were likely experiencing forms of stigma (Earnshaw & Chaudoir, 2009; Quinn & Chaudoir, 2009; Quinn & Earnshaw, 2013; Van Brakel, 2006). Both my personal and work experiences led to my interest in the psychological impact of physical health conditions, particularly in young people. This matched well with the interest of my supervisor, who was interested in stigma experiences in CYP in paediatric populations.

Despite the importance of the research topic, there were few empirical studies or reviews specifically looking at the stigma experiences of CYP with health conditions. Those that did, predominantly sampled CYP with epilepsy (Benson et al., 2015b). While this was useful on the one hand, as it created a niche for our project, on the other, it created a wider base from which to develop the project. When looking back at initial project meetings and proposal drafts for our empirical research, there were many ideas and variables that could have been included. However, this created confusion and uncertainty about how to focus the project. As such, during the initial research proposal, my colleague and I conducted a large systematic scoping search to narrow our potential research questions and to develop a robust rationale for the variables we selected to be most important. This was an incredibly useful process for two reasons. Firstly, there were several variables, for example mental health stigma, which conceptually and anecdotally sounded valid to explore in CYP with physical health conditions. However, when we reviewed the evidence there was not enough research to build a current rationale around these variables in this population. Secondly, this was the first task that my colleague and I collaborated together on and we were able to observe each other's working style, develop clear communication and build a good relationship, which proved extremely valuable going forward.

2.2 Understanding the experiences of CYP with physical health conditions

My experiences with physical health conditions had led me to develop a fairly negative and narrow narrative about how those living with these conditions would feel. Also, the field of research is predominately focused on the difficulties that CYP and adults with health conditions face; for example, the higher risk of developing poorer psychological and physical wellbeing (Gamwell et al., 2018; Hysing et al., 2007) and being victims of bullying and social exclusion (Lambert & Keogh, 2015). Therefore, I assumed that in both my systematic review and in my empirical paper, my findings would reflect examples of CYP feeling highly stigmatised and having difficult feelings related to their physical health conditions. In my systematic review, the findings were consistent with stories of difficulty and common themes suggested that most CYP reported stigmatising experiences and many felt distressed because of their health condition (Benson et al., 2015a; Kirk & Hinton, 2019; Wo et al., 2018). Similarly, in the empirical study some CYP scored highly on the questionnaires measuring self-stigma and illness attitudes and some CYP and their caregivers during the data collection, shared anecdotal stories of teasing or feeling distressed because of their condition.

However, there were also positive stories captured in the systematic review where some CYP reported that differences due to their health condition did not make them abnormal (MacLeod, 2009) and others described times that disclosing their condition had improved relationships with close friends (Kirk & Hinton, 2019; MacLeod, 2009). During the data collection for the empirical study, many CYP and their families also described that their health condition was important, but it was just something that they managed and there were stories of strength and resilience. Although the aims of my systematic review and empirical paper were based on the research literature, they were also shaped by my assumptions and judgements of CYP living with a health condition. Before the research, I viewed adults and CYP with health conditions as 'suffering' with their conditions. I was motivated by a need to contribute something to the literature that would highlight this suffering, albeit in a well-meaning way, to raise awareness amongst the clinical and medical communities. Through completing the research, I have developed a much richer narrative of those living with health conditions. Although there is evidence to suggest that CYP are likely to experience distress in the form of stigmatisation, negative illness attitudes and emotional and peer problems, this is not the only story. I believe being able to think more widely about the experiences of those with health conditions will help me as both personally and professionally as a clinician. For example; in clinical practice when working and supporting individuals and communities who feel stigmatised, I hope to be more aware of my own implicit judgements and assumptions and reflect on how this may impact assessments, formulations or interventions. Furthermore, if I were to pursue more research in this area, I would try and encapsulate the variety of experiences of CYP with health conditions through the use of different questionnaires or exploratory qualitative methods.

2.3 Managing Difficulties

The empirical study was a joint project with a fellow trainee. Developing a good working relationship was invaluable to the project. We were able to learn about each other's strengths and limitations and how to capitalise on these so that we shared tasks appropriately and matched to each other's skill set. We were also able to bring different perspectives and ideas which helped to pre-empt, and problem solve difficulties that appeared along the way. In particular, I noticed that having accountability for someone else helped to motivate me, stay organised and to complete tasks in a timely manner.

Evidence suggests that emotional and peer support helps to manage stress responses and enhance learning experiences for trainee clinical psychologists during training (Kuyken et al., 2003; Nel et al., 2012). This was something I was able to relate to. Having a fellow trainee to reflect, seek support and problem solve with during times of difficulty, helped me to manage my own anxieties and stress throughout the project. For example, during the data collection process, I found it particularly challenging when CYP answered the questionnaires in such a way that indicated they felt distressed about their physical health condition. Although I ensured that they received an appropriate debrief and made the CYP and family aware of the relevant support services on offer, my clinical urge in that moment was to offer support or guidance to the CYP and their family. A review found that during research, clinicians struggle to exclusively assume a non-clinical research identity and that many clinicians feel drawn to interact with research participants as if they were clinical patients (Hay-Smith et al., 2016). Being able to receive peer support from someone else who was also actively part of the data collection provided a useful space for me to acknowledge my feelings of sadness, frustration and sense of helplessness but also reflect on the role and boundaries of being a researcher.

Another difficultly occurred at the beginning of March when the onset of the COVID-19 in the UK meant that we needed to change our recruitment strategy. The pandemic itself elicited many anxieties for me personally; I had concerns about family members, my final training placement was uncertain, and the data collection added an additional stress. We took steps quickly to adapt to remote data collection and having a fellow trainee to develop this new plan with helped me to feel less overwhelmed. In addition, we were both able to reflect, together and with our supervisor, about the potential ethical dilemmas of collecting data during the pandemic and thought about the immediate needs of the CYP and their families, the impact of lockdown, home-schooling and health concerns. As such, we temporarily paused remote data collection for a short time to provide space for families to adjust to changes, rather than ploughing ahead and potentially adding further demands on caregiver schedules. Although we had to extend our data collection timeframe, reducing the time planned for data analysis and write up, making this decision together with my fellow trainee and supervisor helped to put the very difficult situation into perspective, and put the needs of the CYP and their families first.

3. Reflections and Critiques on Methodology and Data Collection

3.1 Choice of Methodology

From completing systematic searches of the literature, there was a prevalence of qualitative methodology and it appeared that using quantitative methods for the empirical paper could further the understanding of the experiences of CYP with health condition. Furthermore, quantitative methods were appropriate for the research questions. However, I am aware from my previous academic work, that I have affinity for quantitative methodology and feel more confident analysing quantitative data. Evidence suggests that many researchers have a bias towards either qualitative or quantitative methodology, based on prior knowledge and confidence, and can find it difficult to shift perspectives (Bryman, 2007; Mahoney & Goertz, 2006). During the systematic review, I became much more attuned to reading and critically appraising qualitative studies. I found some of the qualitative papers incredibly moving and they enhanced my understanding of CYP with health conditions, beyond that of the quantitative papers I read. I believe that applying a quantitative methodology to the empirical paper has made valuable contributions to the research field. However, if I were to approach similar concepts again, I would consider using qualitative or a mixed methods approach to gain more insight into the experiences of CYP with health conditions. I think this could be particularly useful when considering the visibility of a physical health condition and understanding more about the different experiences CYP may have with visible or less visible conditions.

The empirical study indicated that the CYP with visible differences had significantly poorer illness attitudes and greater emotional and peer difficulties. It would be valuable to explore in greater depth how CYP view the visibility of their condition and what role it plays in how they feel towards their health condition and how this impacts their emotional feelings and peer relationships. This is not an area well researched currently in the literature. By understanding more about these links, it could enhance the effectiveness of psychological interventions and support CYP to develop acceptance and more positive feelings to living with a physical health condition.

3.2 Questionnaires and Measures

Following the systematic search of the literature, the measures employed in the empirical study were selected based upon their prior use with CYP with health conditions and the appropriateness of the language used for CYP between 8-14 years old. However, due to the limited research comparing visible and less visible conditions and CYP with physical health conditions more generally, some of the questionnaires or methods of measurement proved difficult to select.

An appropriate measure of concealment was hard to source. Many studies exploring experiences of concealment in those with health conditions used language that was not accessible to CYP (Quinn et al., 2017), or others who did sample CYP with physical health conditions developed their own questionnaires specific to their study (Benson et al., 2016). After discussions with my supervisor and fellow trainee, the measure of concealment used was adapted from the secrecy sub-scale, part of a child and adolescent mental health stigma scale, the Paediatric Self-Stigmatisation Scale (PeadS) (Kaushik et al., 2017). This was chosen because it was suitable for CYP between 8-12 years, the questions could be easily changed to accommodate views towards physical health conditions, and it was only seven questions. Findings from the systematic review later highlighted another strength of the questionnaire, in that it asks about both concealment related to the condition and concealment regarding treatment for the condition, both of which CYP describe as concealment related concerns (Benson et al., 2015a; Lewis & Parsons, 2008;

Olsson et al., 2009). However, there were several limitations. Firstly, the scale had not previously been used with CYP with physical health conditions and some evidence suggests there can be differences in how CYP view and report mental health conditions and physical health conditions (Butler et al., 2015; Corrigan et al., 2005). In addition, the first item of the secrecy scale used was phrased in a complicated way and also asked from a third person perspective. This was not identified as being too difficult to answer by seven CYP who participated in the Patient and Public Involvement & Engagement (PPI-E). However, when administering the measures, both face to face and remotely, some CYP found the item confusing and it is possible that those who did not seek help may not have answered the item the way they intended. Future research could consider developing a validated measure based on the qualities of existing questionnaires to assess concealment practices in CYP which could be adapted easily to suit different physical health conditions.

As already discussed in the empirical paper, the visibility of a condition is complex (Rumsey & Harcourt, 2007), and future research could consider using a scale measurement to capture some of this complexity. In addition, the aspect of visibility may also interact with specific conditions and is evidenced in the empirical paper where findings suggested that there can be an association between visibility and CYP from dermatology clinics. Another quantitative way to understand the impact of visibility and how this impact may be similar or different in certain conditions could be to present CYP a series of statements which represent specific times their condition may be visible to others. Some CYP report their condition is only visible during flare ups (Kirk & Hinton, 2019), following the sudden onset of symptoms (Benson et al., 2015a), or following specific requests in front of others (Olsson et al., 2009). For example, CYP with sickle cell disorder reported requesting permission to leave a classroom for a drink of water to help manage their pain (Dyson et al., 2010). This may help to understand both condition specific factors and situational factors which can lead to the condition being more or less visible, and how such factors impact how CYP feel about their condition and their psychosocial functioning. Similar statements have been trialled in questionnaires measuring disclosure attitudes in adults with a range of physical health, mental health and other concealable identities (Quinn et al., 2017). A similar framework could be used to measure aspects of visibility in CYP with visible and less visible physical health conditions.

3.3 Data Collection

The findings of the study cannot be generalisable to Black, Asian and other minority ethnic groups due to the majority of CYP and their families being from White or White British backgrounds. Evidence indicates that physical health stigma is likely to intersect with other stigmatised identities, such as gender, sexual orientation, ethnicity, religion and culture (Remedios & Snyder, 2015; Stangl et al., 2019; Turan et al., 2019). Therefore, there is a need to recruit more diverse samples to understand how physical health stigma, concealment, illness attitudes and disease specific factors such as visibility may have different effects across ethnic groups. Additionally, there are systemic barriers and clear examples of health inequalities that negatively affect Black, Asian and minority ethnic groups and it is even more essential that research in healthcare populations are representative of diverse groups. On reflection, there are many ways which we could have made our recruitment methods more inclusive. Firstly, we could have discussed the role of our own cultural background as researchers and how this may influence the research and how we may have been perceived by possible participants. We then could have met with service user groups, including CYP, their families and medical staff teams to discuss their views on the research, challenges they foresaw, and ideas they had towards creating a more inclusive recruitment strategy. We could have had greater presence in the clinics and discussed the study informally with families first to develop relationships. We also could have discussed ways to use interpreters with the medical team. The study has shown me that as a researcher or a clinician I need to work much harder to ensure that the work I produce represents those from diverse groups.

4. Conclusions

This project has provided me with a huge insight into the range of experiences of children and young people with physical health conditions. Particularly how different types of stigma and the visibility of a physical condition can contribute towards how CYP feel about their health condition and how they feel about keeping it hidden from others. The project also highlights the need for better awareness in clinical settings about some of the difficulties CYP with physical health conditions face and the need for interventions to specifically target stigma and condition specific factors such as visibility. Hopefully this critical appraisal can help future clinicians or researchers think about ways to address some of the issues discussed and to take forward new ideas to better support CYP living with physical health conditions.

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Weiss, M. G., Ramakrishna, J., & Somma, D. (2006). Health-related stigma: rethinking concepts and interventions. *Psychology, Health & Medicine, 11*(3), 277-287. doi: 10.1080/13548500600595053

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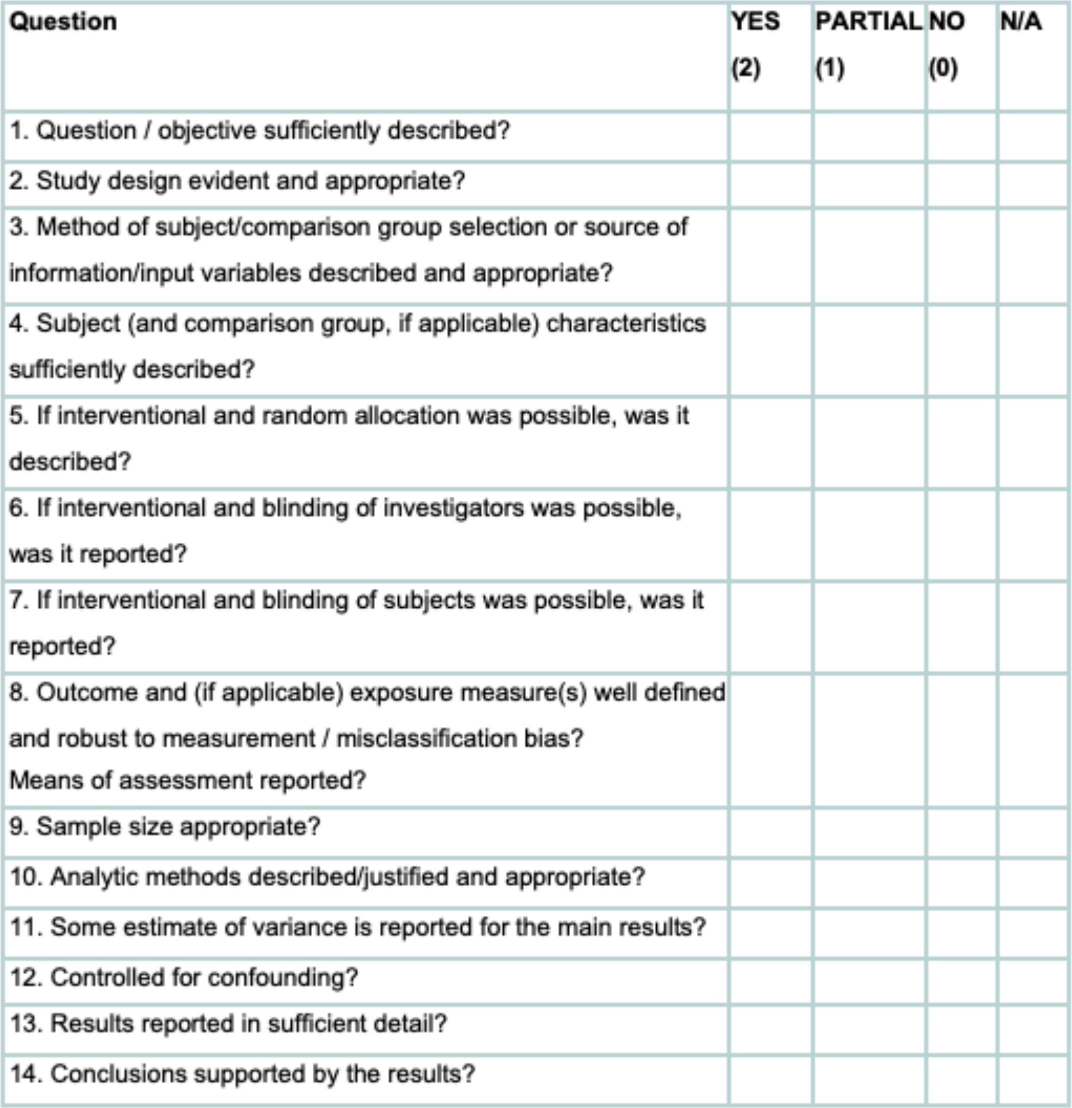
Appendices

Appendix A: Critical Appraisal Tool - Quantitative Studies

Standard Quality Assessment Criteria for Evaluating Primary Research Papers from a Variety of Fields (Kmet et al., 2004)

14 items were scored depending on the degree to which the specific criteria were met (“yes” = 2, “partial” = 1, “no” = 0). Items not applicable to a particular study design were marked “n/a” and were excluded from the calculation of the summary score. A summary score was calculated for each paper by summing the total score obtained across relevant items and dividing by the total possible score (i.e.: 28 – (number of “n/a” x 2))

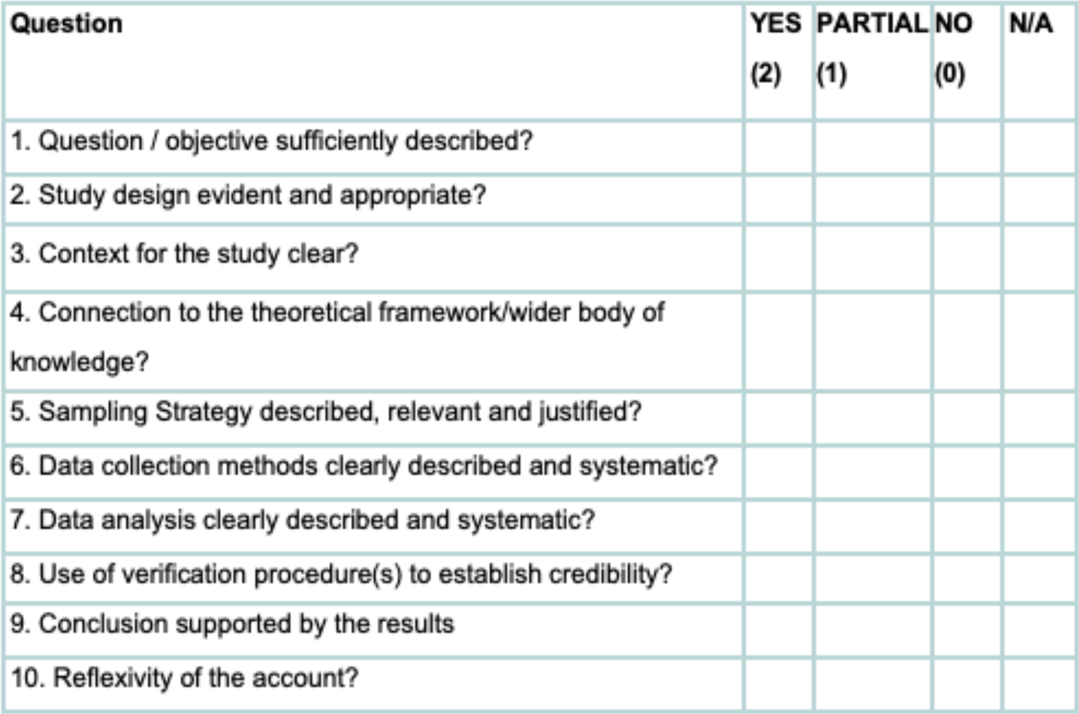


Question	YES (2)	PARTIAL (1)	NO (0)	N/A
1. Question / objective sufficiently described?				
2. Study design evident and appropriate?				
3. Method of subject/comparison group selection or source of information/input variables described and appropriate?				
4. Subject (and comparison group, if applicable) characteristics sufficiently described?				
5. If interventional and random allocation was possible, was it described?				
6. If interventional and blinding of investigators was possible, was it reported?				
7. If interventional and blinding of subjects was possible, was it reported?				
8. Outcome and (if applicable) exposure measure(s) well defined and robust to measurement / misclassification bias? Means of assessment reported?				
9. Sample size appropriate?				
10. Analytic methods described/justified and appropriate?				
11. Some estimate of variance is reported for the main results?				
12. Controlled for confounding?				
13. Results reported in sufficient detail?				
14. Conclusions supported by the results?				

Appendix B: Critical Appraisal Tool - Qualitative Studies

Standard Quality Assessment Criteria for Evaluating Primary Research Papers from a Variety of Fields (Kmet et al., 2004)

10 items were scored depending on the degree to which the specific criteria were met (“yes” = 2, “partial” = 1, “no” = 0). Items not applicable to a particular study design were marked “n/a” and were excluded from the calculation of the summary score. A summary score was calculated for each paper by summing the total score obtained across relevant items and dividing by the total possible score (i.e.: 28 – (number of “n/a” x 2))

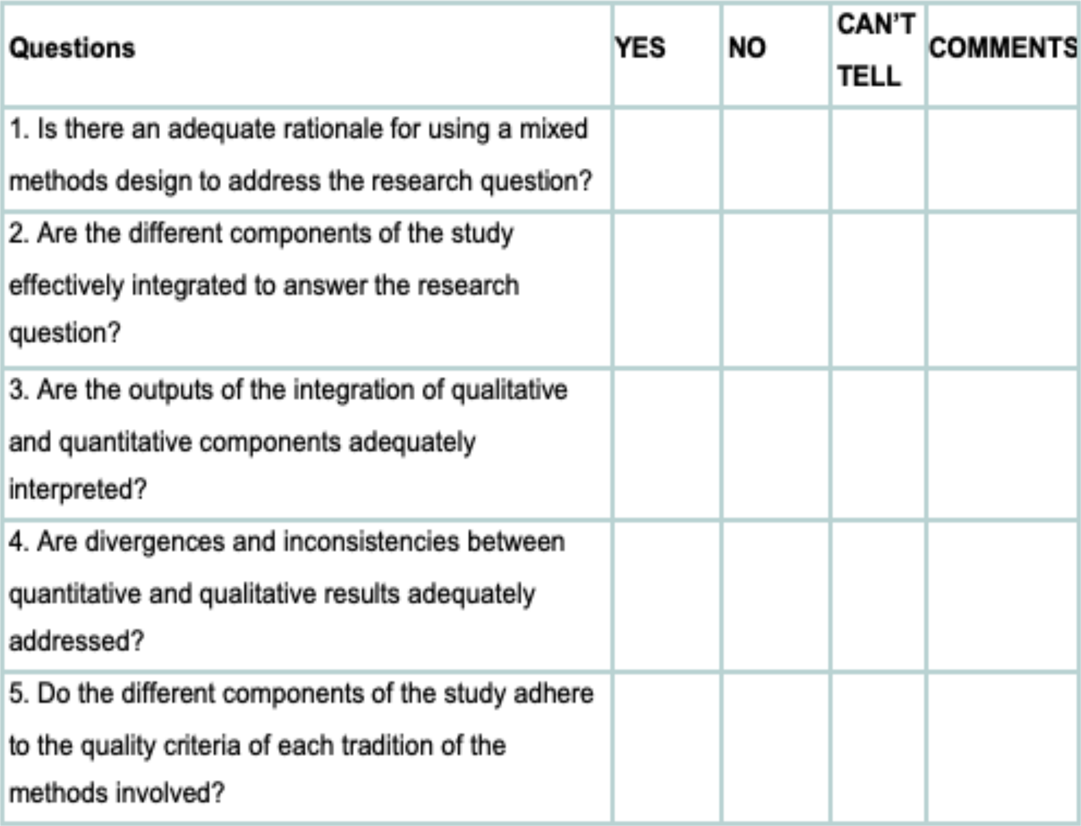


Question	**YES (2)**	**PARTIAL (1)**	**NO (0)**	**N/A**
1. Question / objective sufficiently described?				
2. Study design evident and appropriate?				
3. Context for the study clear?				
4. Connection to the theoretical framework/wider body of knowledge?				
5. Sampling Strategy described, relevant and justified?				
6. Data collection methods clearly described and systematic?				
7. Data analysis clearly described and systematic?				
8. Use of verification procedure(s) to establish credibility?				
9. Conclusion supported by the results				
10. Reflexivity of the account?				

Appendix C: Critical Appraisal Tool – Mixed Methods Study

Mixed Methods Appraisal Tool (MMAT) (Hong et al., 2018)

The MMAT does not calculate an overall score, it is advised to perform a more detailed presentation of the ratings of each criterion to better inform the quality of the included



Questions	**YES**	**NO**	**CAN'T TELL**	**COMMENTS**
1. Is there an adequate rationale for using a mixed methods design to address the research question?				
2. Are the different components of the study effectively integrated to answer the research question?				
3. Are the outputs of the integration of qualitative and quantitative components adequately interpreted?				
4. Are divergences and inconsistencies between quantitative and qualitative results adequately addressed?				
5. Do the different components of the study adhere to the quality criteria of each tradition of the methods involved?				

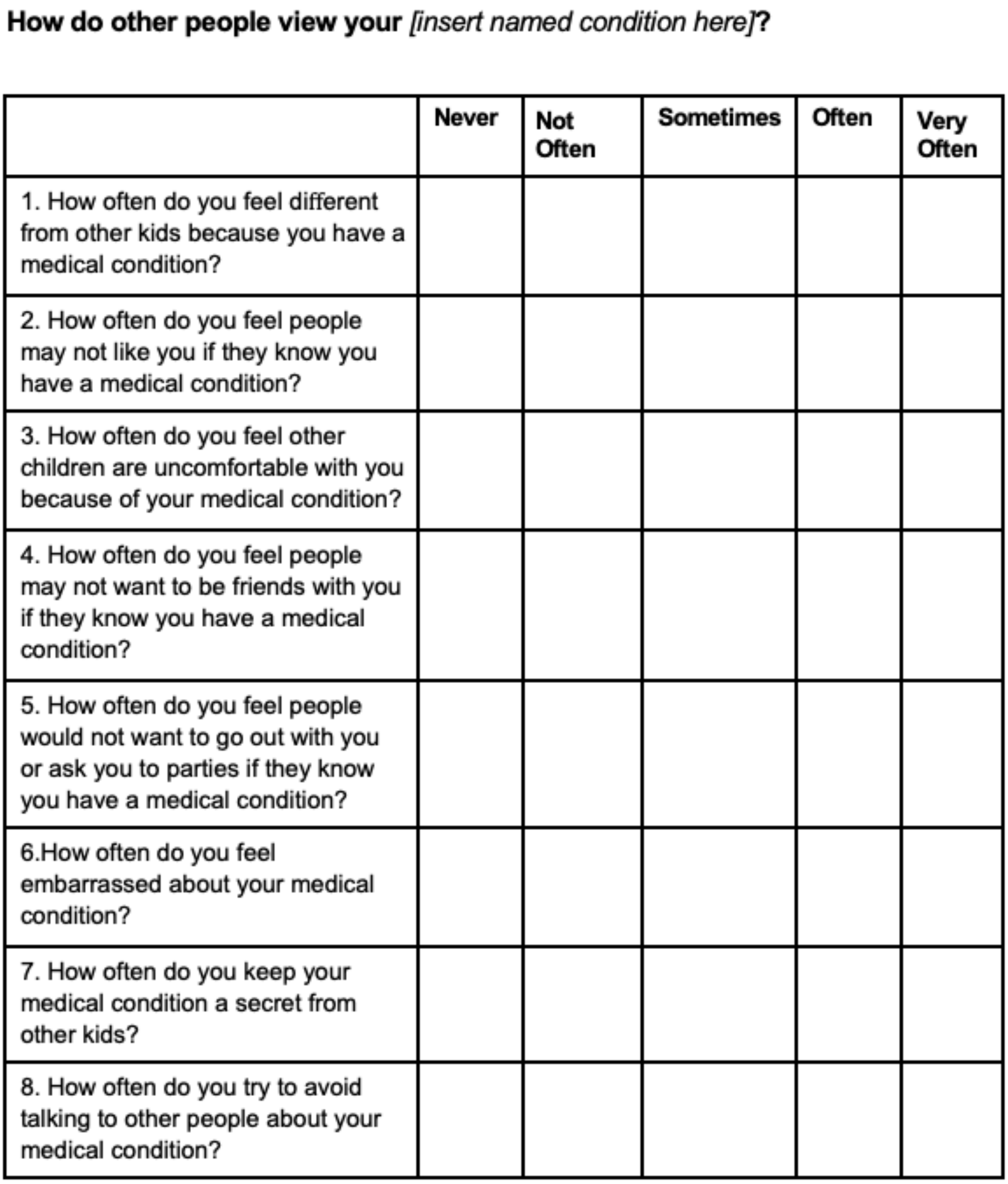
Appendix D: Joint Thesis Declaration

This was a joint project working in collaboration with Claire Hackford, who was exploring stigma and psychosocial difficulties of CYP with physical health conditions, specifically the associations between CYP and caregivers (Hackford, 2020).

Systematic Review: Claire Hackford acted as the second-rater and critically appraised eight of the studies included in the systemic review.

Empirical Paper: Both trainees conducting initial scoping searches together to understand the literature and develop meaningful research questions. The recruitment strategy was jointly planned and adapted, with each trainee having an equal role in the data collection. The subsequent analysis and write up of the findings were carried out independently.

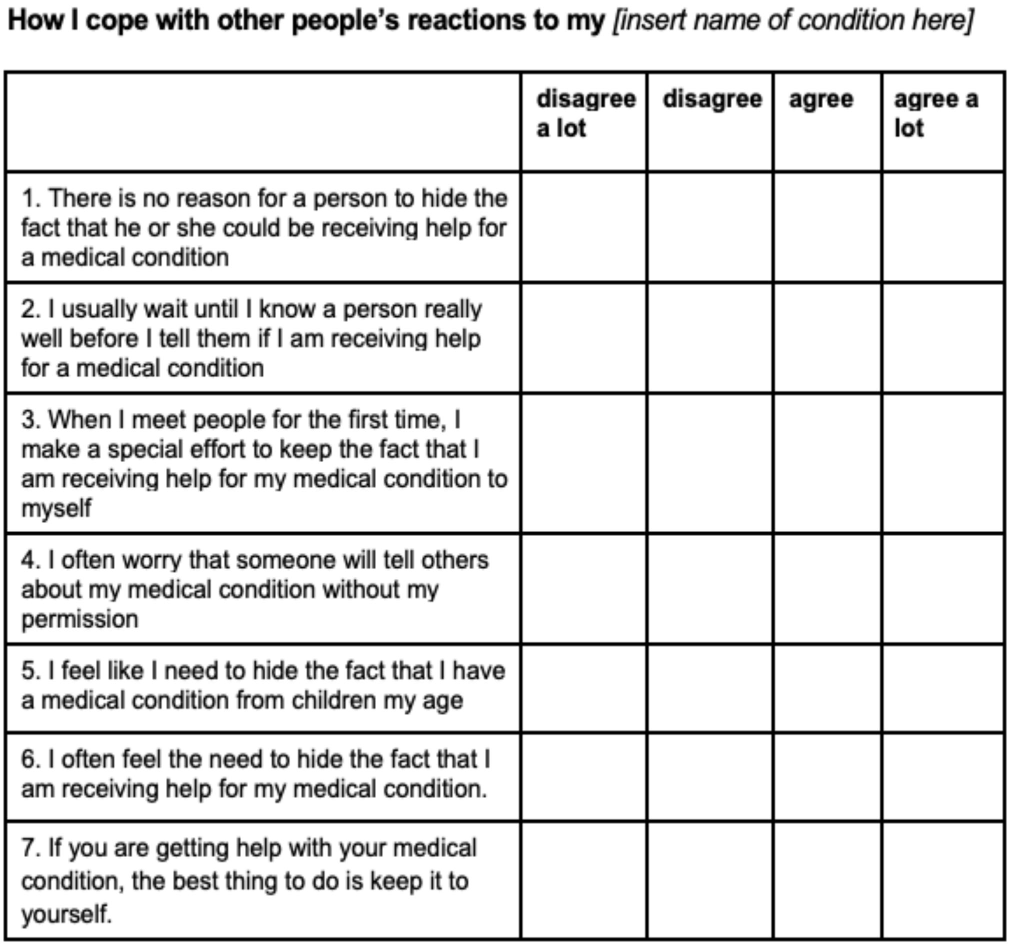
Appendix E: Self-Stigma Questionnaire (CYP)



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?					

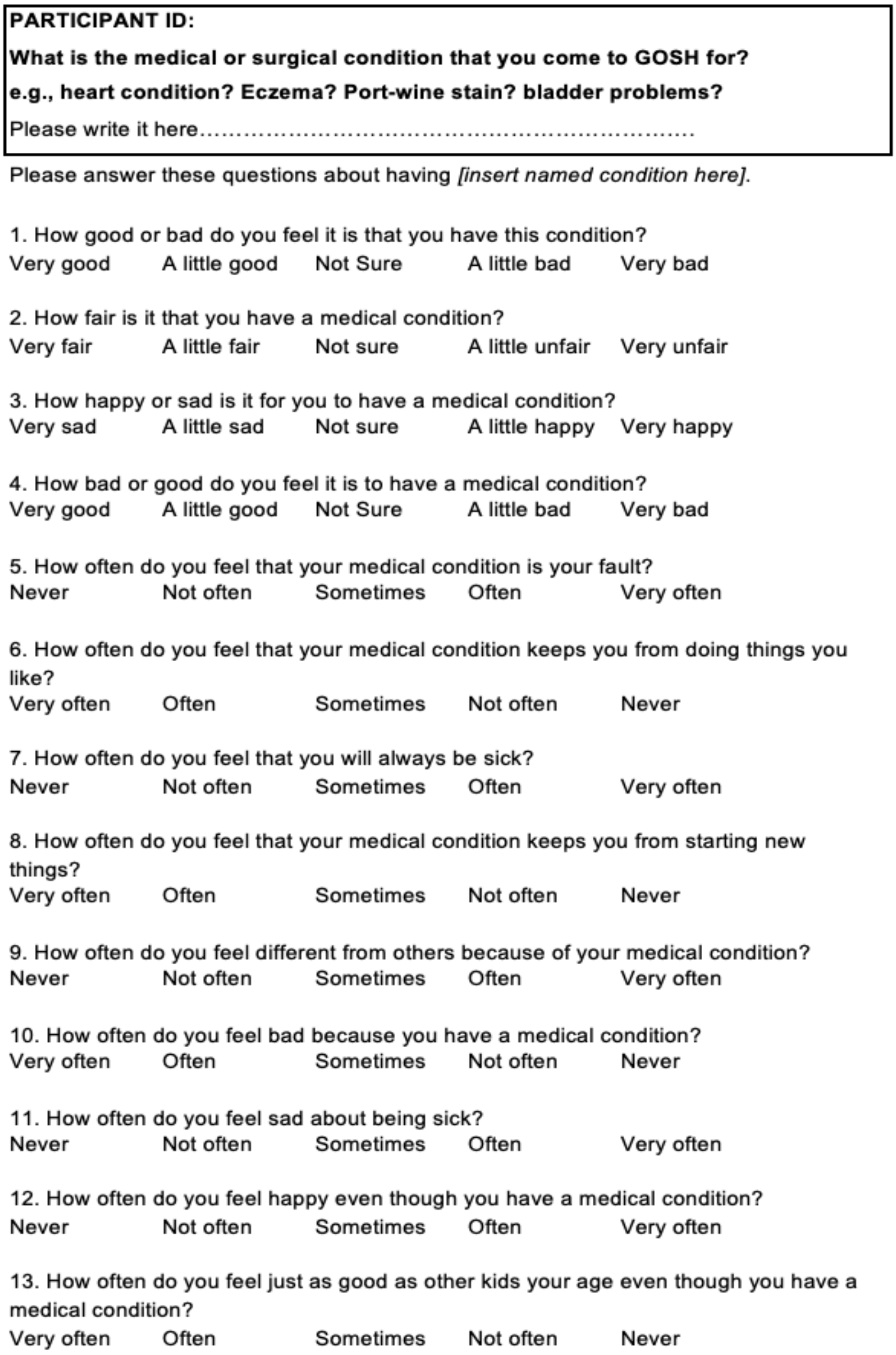
Appendix F: Concealment Questionnaire (CYP)



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 G: Illness Attitudes Questionnaire (CYP)



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

Appendix H: Demographic and Medical Profile Questionnaire (Parents)

BACKGROUND INFORMATION

PARTICIPANT ID:

1) Child's age:

Age: …… years ……months

2) Please state your child's gender identity:

……………………………………………

3) School year:

☐ Year 3 ☐ Year 4 ☐ Year 5
☐ Year 6 ☐ Year 7 ☐ Year 8
☐ Year 9 ☐ Year 10 ☐ 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*

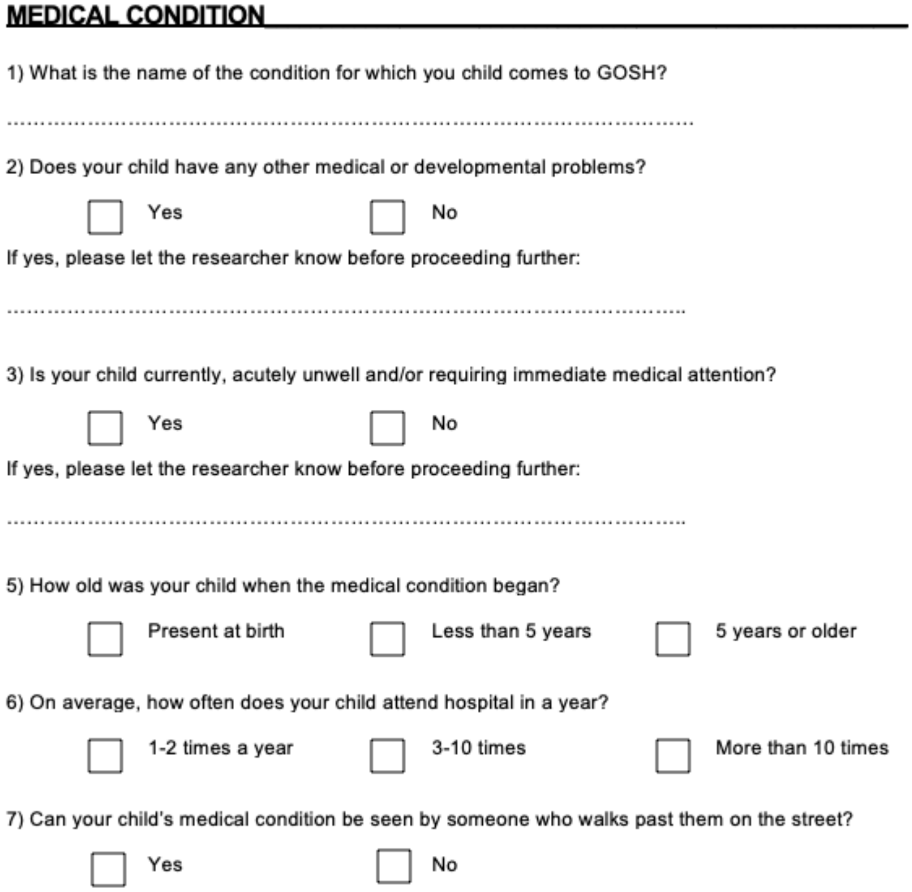
C Asian/Asian British
☐ Indian
☐ Pakistani
☐ Bangladeshi
☐ Chinese
☐ Any Other Asian background, *please write in*

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
☐

Appendix H (cont.): Demographic and Medical Profile Questionnaire (Parents)



MEDICAL CONDITION

1) What is the name of the condition for which you 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

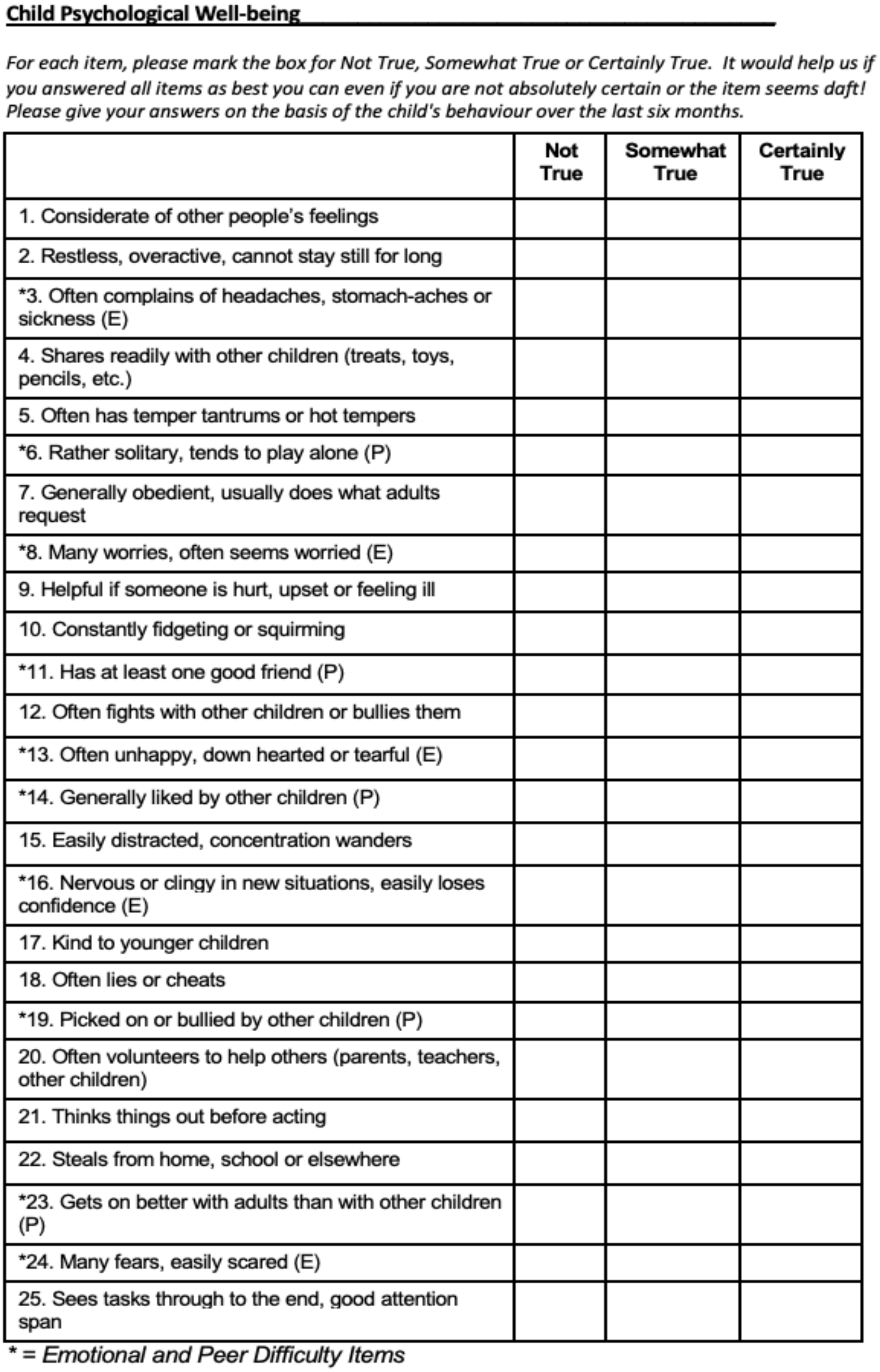
6) On average, how often does your child attend hospital in a year?

☐ 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

Appendix I: Emotional and Peer Difficulties Questionnaire



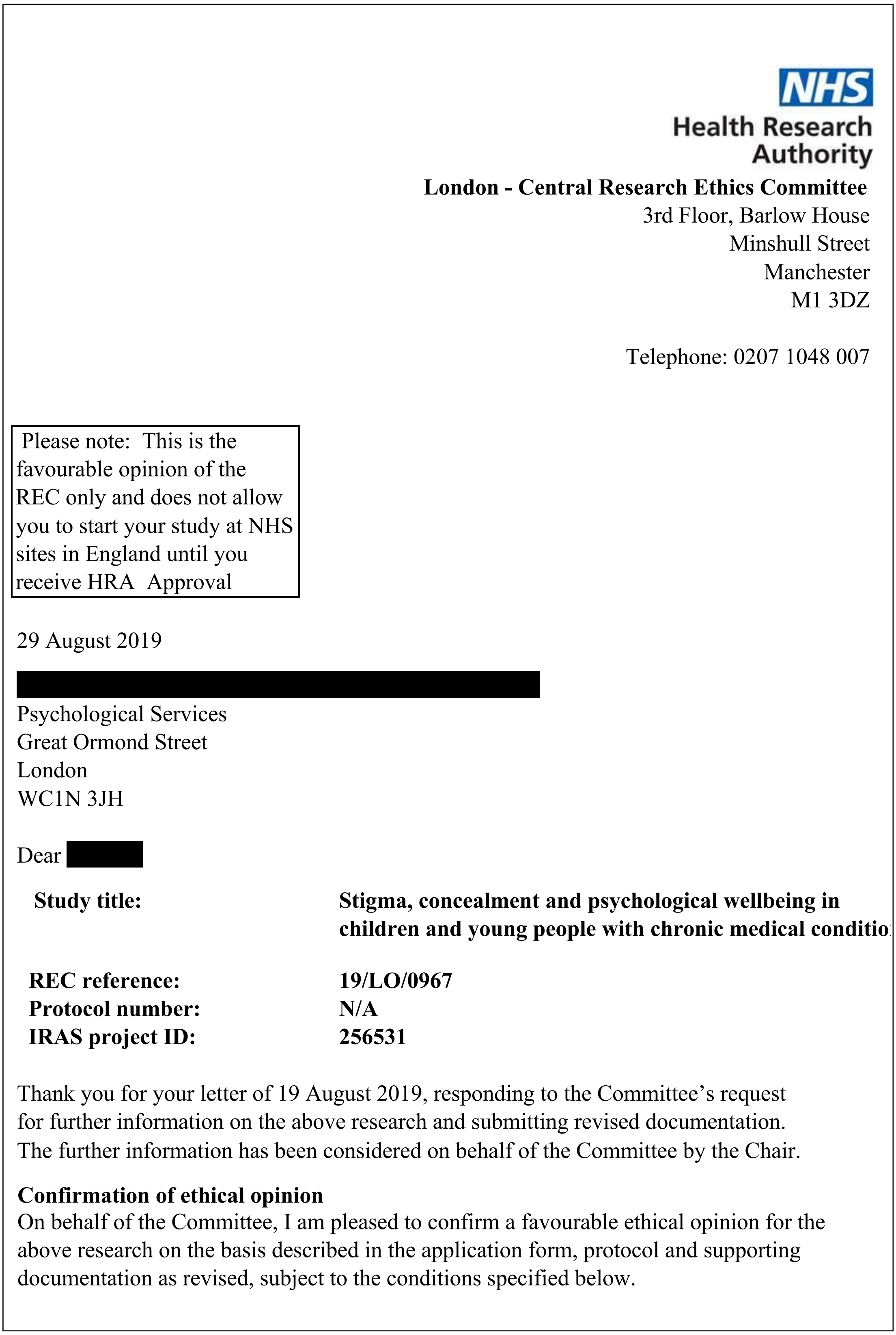
Child Psychological Well-being

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 (E)			
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 (P)			
7. Generally obedient, usually does what adults request			
*8. Many worries, often seems worried (E)			
9. Helpful if someone is hurt, upset or feeling ill			
10. Constantly fidgeting or squirming			
*11. Has at least one good friend (P)			
12. Often fights with other children or bullies them			
*13. Often unhappy, down hearted or tearful (E)			
*14. Generally liked by other children (P)			
15. Easily distracted, concentration wanders			
*16. Nervous or clingy in new situations, easily loses confidence (E)			
17. Kind to younger children			
18. Often lies or cheats			
*19. Picked on or bullied by other children (P)			
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 (P)			
*24. Many fears, easily scared (E)			
25. Sees tasks through to the end, good attention span			

** = Emotional and Peer Difficulty Items*

Appendix J: Ethical approval from the Health Research Authority



NHS
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

[redacted]
Psychological Services
Great Ormond Street
London
WC1N 3JH

Dear [redacted]

Study title:	**Stigma, concealment and psychological wellbeing in children and young people with chronic medical conditio**
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.

Appendix J (cont.): Ethical approval from the Health Research Authority

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

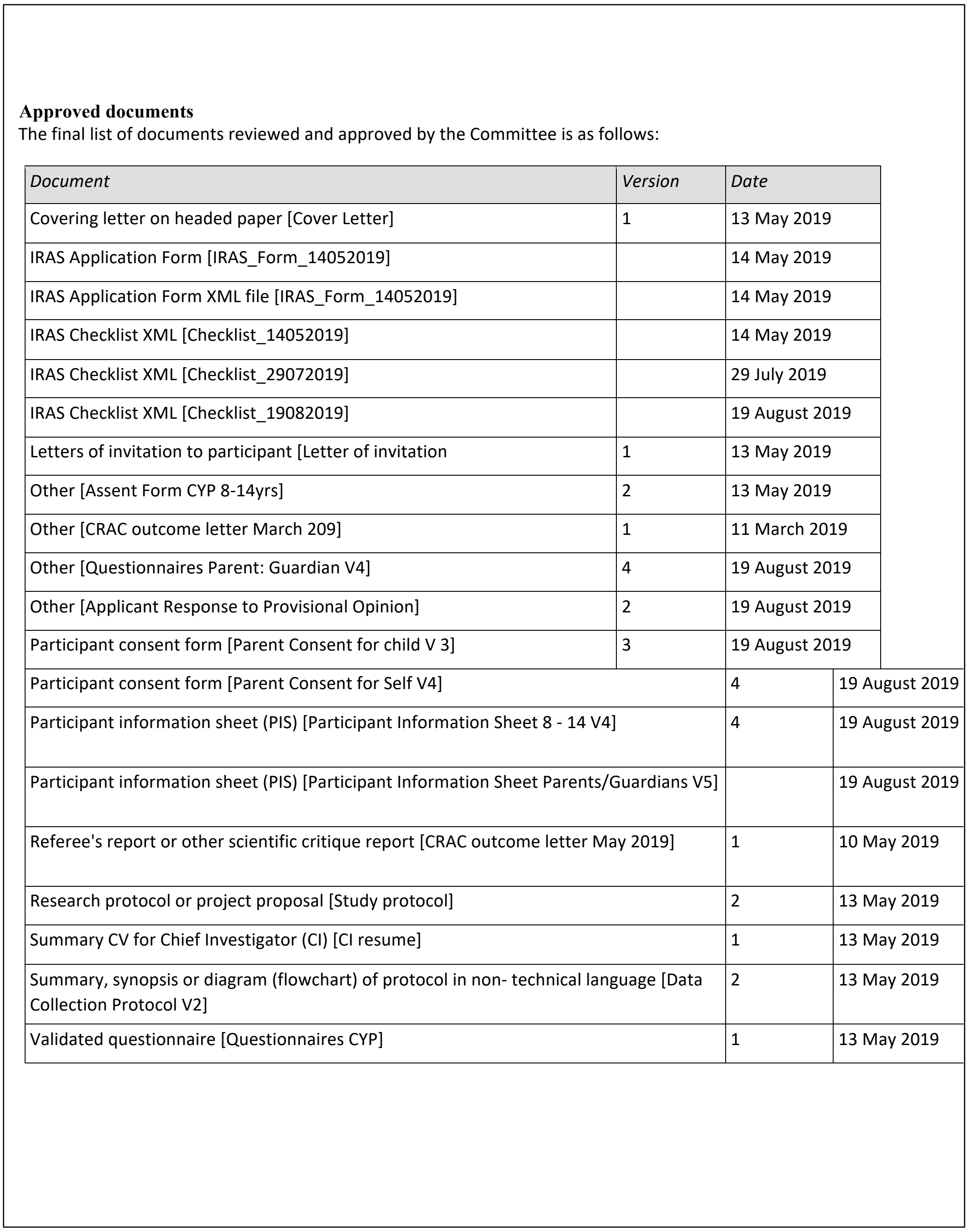
NHS/HSC sites

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.

Appendix J (cont.): Ethical approval from the Health Research Authority



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

Document	*Version*	*Date*
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

[redacted]

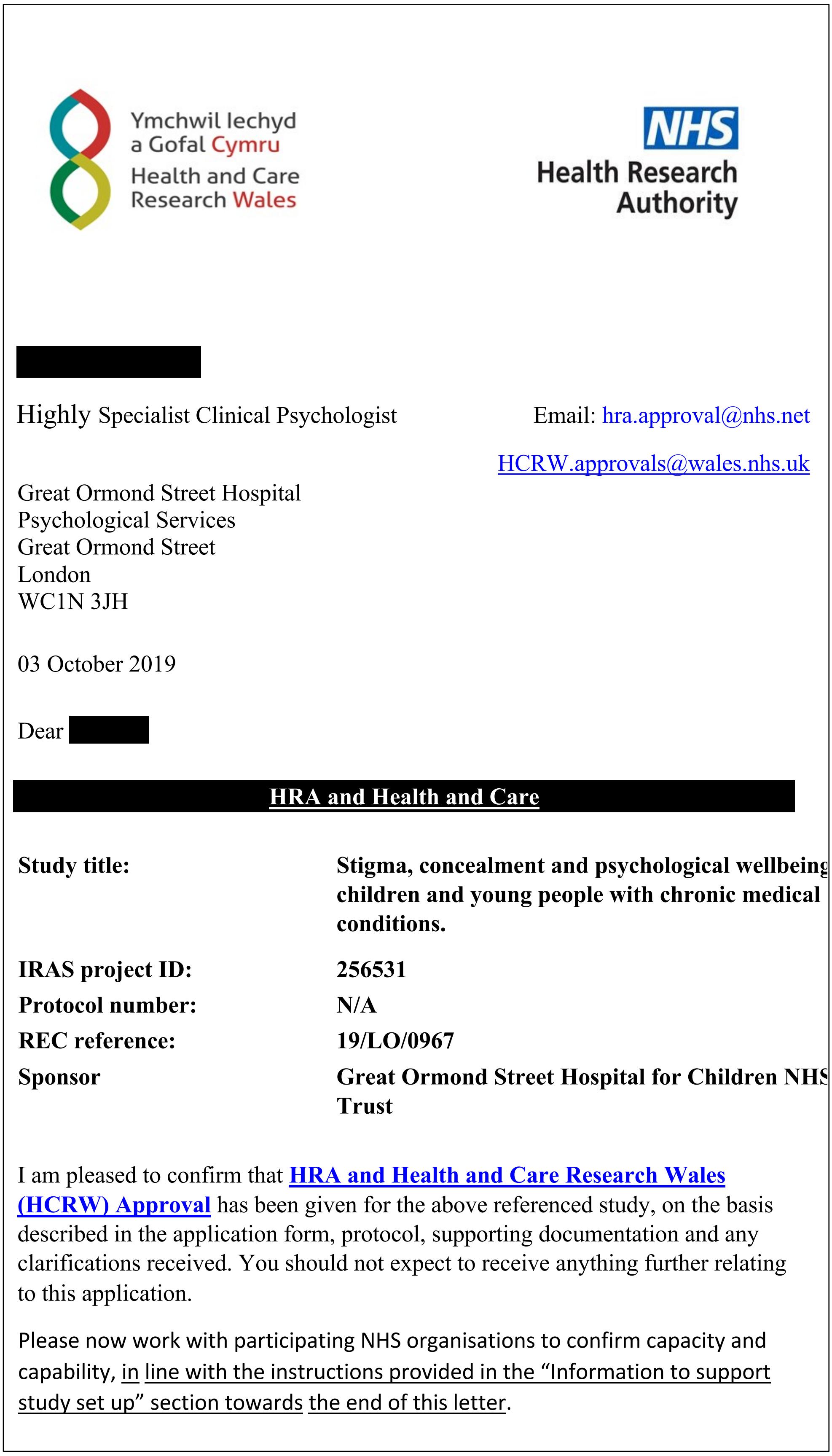
Dr Andrew Hilson Chair

Email: NRESCommittee.London-Central@nhs.net

Enclosure: "After ethical review – guidance for researchers"

Copy to: Ms Vanshree Patel

Appendix J (cont.): Ethical approval from the Health Research Authority



Ymchwil Iechyd
a Gofal Cymru
Health and Care
Research Wales

NHS
Health Research
Authority

[redacted]
Highly Specialist Clinical Psychologist

Email: hra.approval@nhs.net
HCRW.approvals@wales.nhs.uk

Great Ormond Street Hospital
Psychological Services
Great Ormond Street
London
WC1N 3JH

03 October 2019

Dear [redacted]

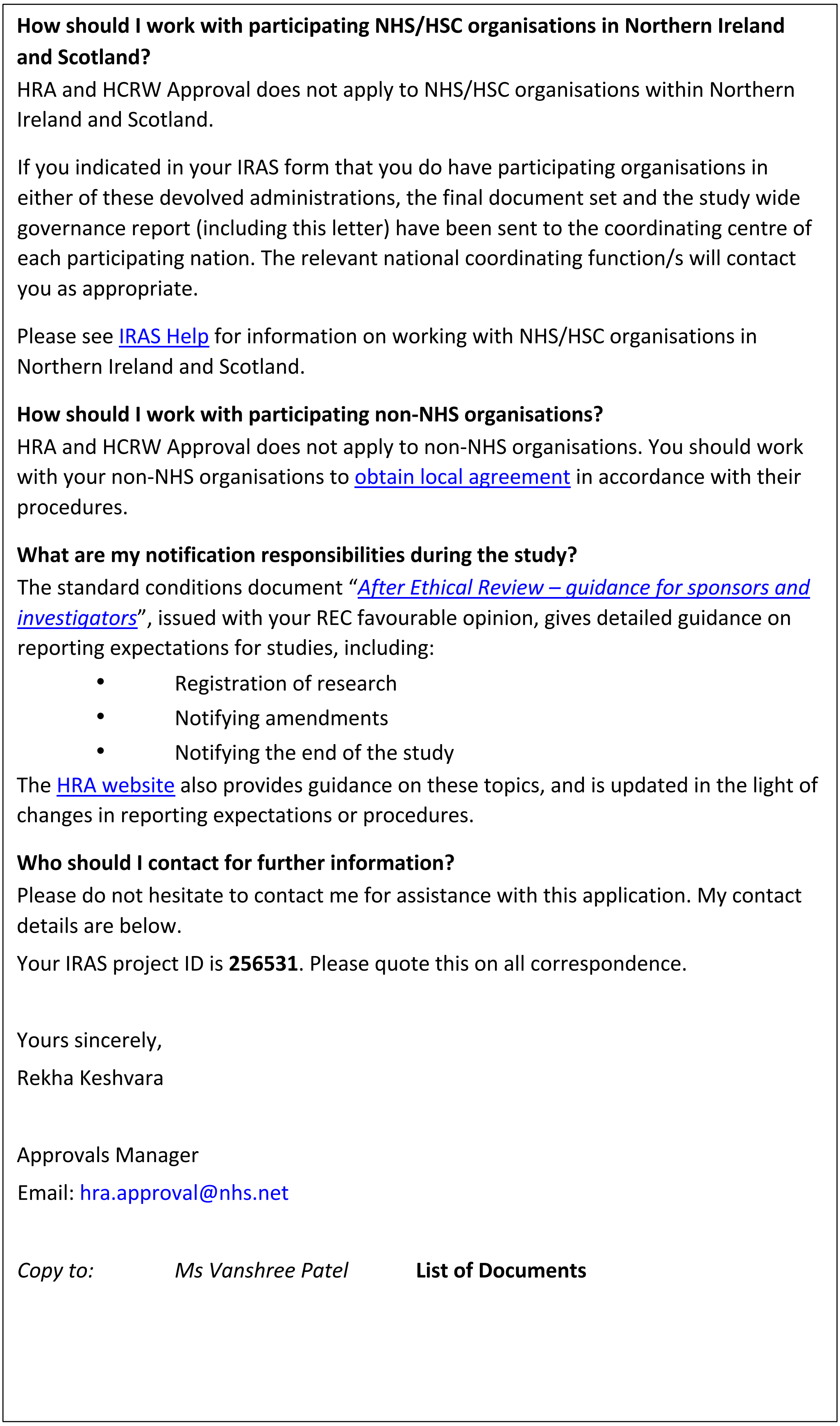
HRA and Health and Care

Study title:	**Stigma, concealment and psychological wellbeing 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.

Appendix J (cont.): Ethical approval from the Health Research Authority



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**

Appendix J (cont.): Ethical approval from the Health Research Authority

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

Document	*Version*	*Date*
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
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IRAS Checklist XML [Checklist_19082019]		19 August 2019
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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
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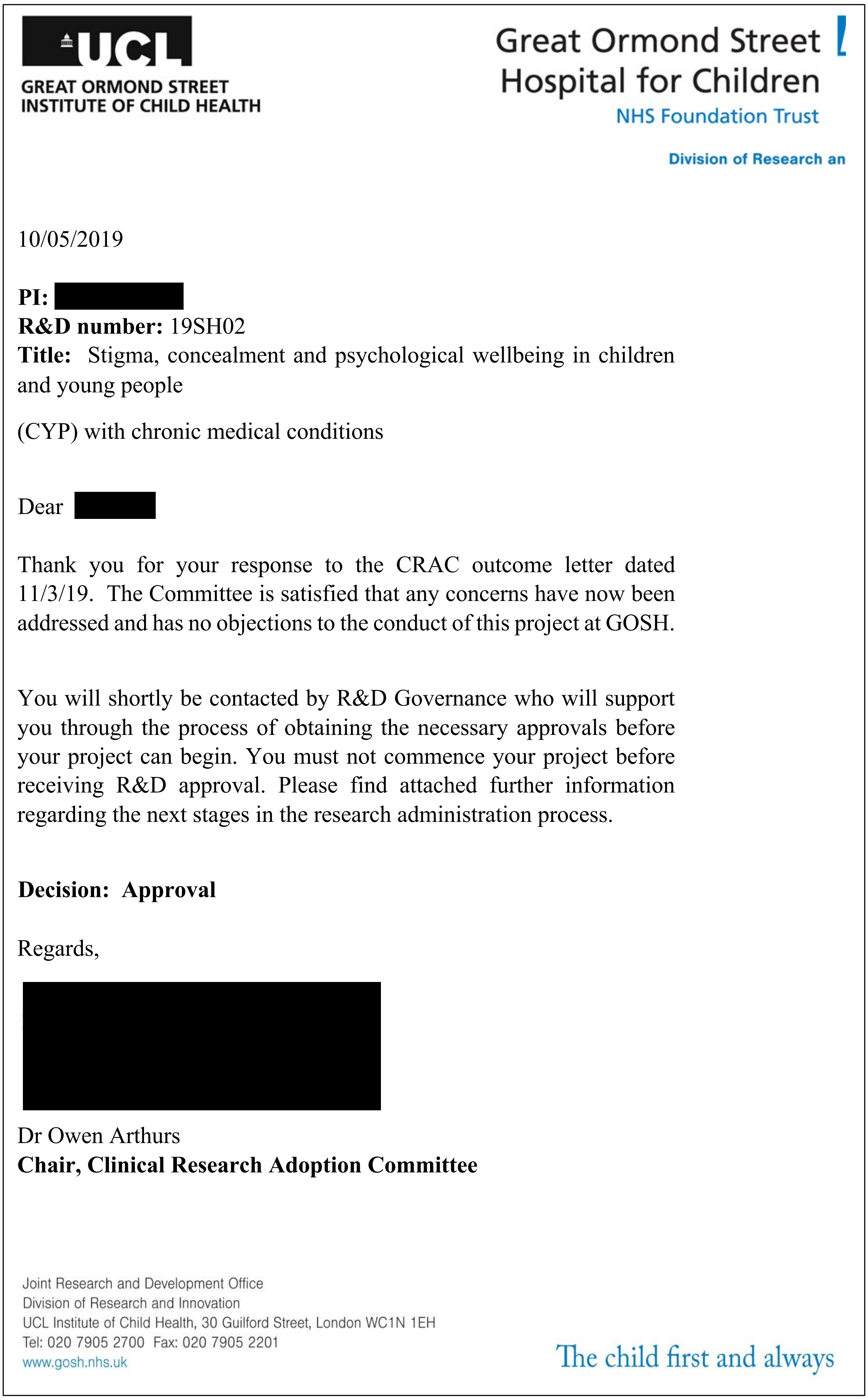
Research protocol or project proposal [Study protocol]	2	13 May 2019
Summary CV for Chief Investigator (CI) [CI resume]	1	13 May 2019
Summary, synopsis or diagram (flowchart) of protocol in non technical language [Data Collection Protocol V2]	2	13 May 2019
Validated questionnaire [Questionnaires CYP]	1	13 May 2019

Appendix K: Ethical approval from the Clinical Research Adoptions Committee at Great Ormond Street Hospital



UCL
GREAT ORMOND STREET
INSTITUTE OF CHILD HEALTH

Great Ormond Street
Hospital for Children
NHS Foundation Trust

Division of Research an

10/05/2019

PI: [redacted]
R&D number: 19SH02
Title: Stigma, concealment and psychological wellbeing in children and young people

(CYP) with chronic medical conditions

Dear [redacted]

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,

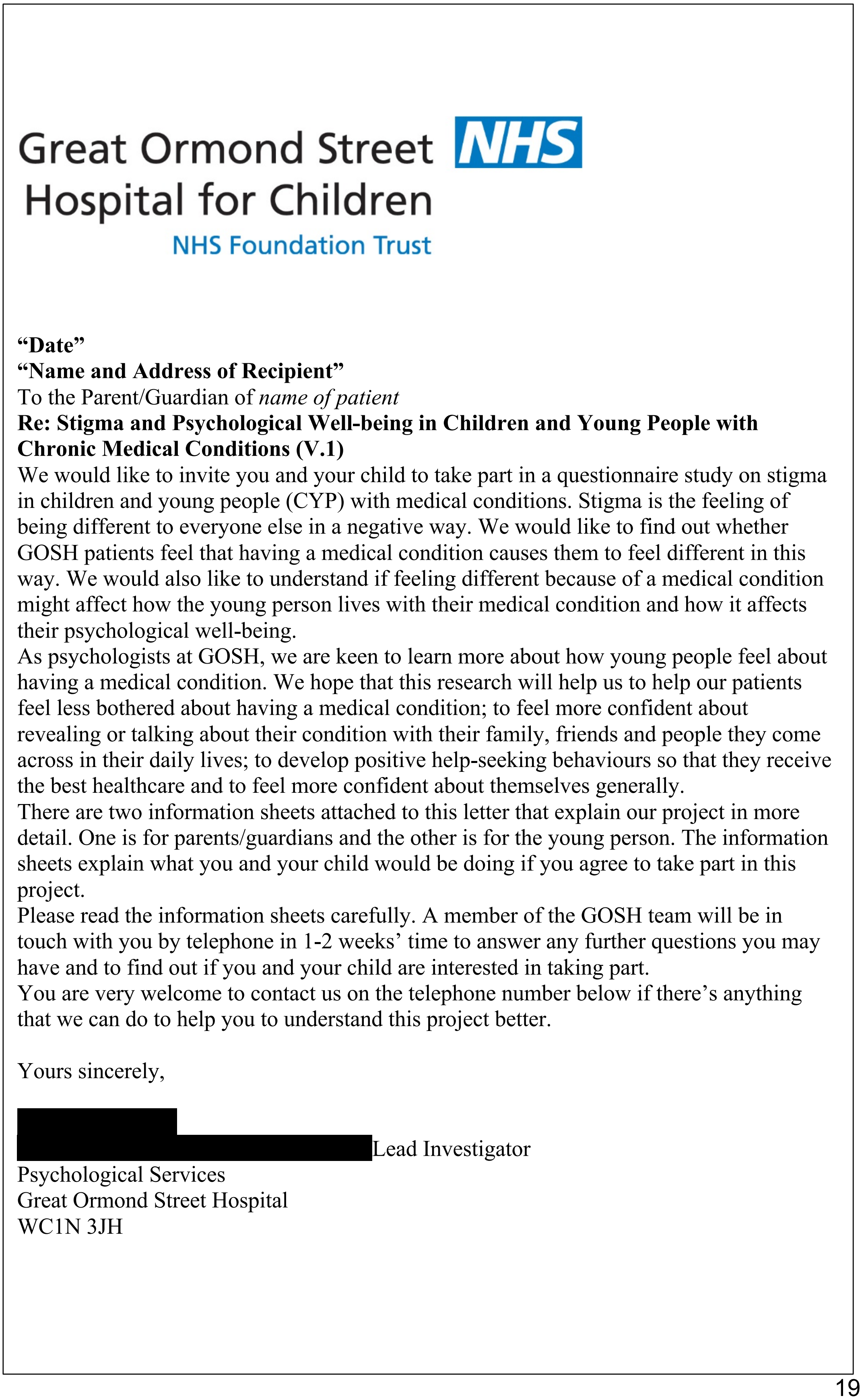
[redacted]

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 L: Letter of Invitation



Great Ormond Street NHS
Hospital for Children
NHS Foundation Trust

"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,

[redacted] Lead Investigator
Psychological Services
Great Ormond Street Hospital
WC1N 3JH

Appendix M: Parent Information Sheet

Participant Information Sheet: Parents/Guardians – Page 1

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.

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Participant Information Sheet: Parents/Guardians – Page 2

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.

Appendix M (cont.): Parent Information Sheet

Participant Information Sheet: Parents/Guardians – Page 3

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:

[redacted]Lead Investigator
Great Ormond Street Hospital
[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
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: 256531), and the Great Ormond Street Hospital Clinical Research Adoptions Committee (Registration No 19SH02).

Participant Information Sheet: Parents/Guardians – Page 4

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 [redacted] Data Protection Officer for Great Ormond Street Hospital, at Your.Data@gosh.nhs.uk

Appendix N: Children & Young People's Information Sheet

Participant Information Sheet: Patients 8-14 years old –Page 1

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.

Appendix N (cont.): Children & Young People's Information Sheet

Participant Information Sheet: Patients 8-14 years old –Page 2

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.

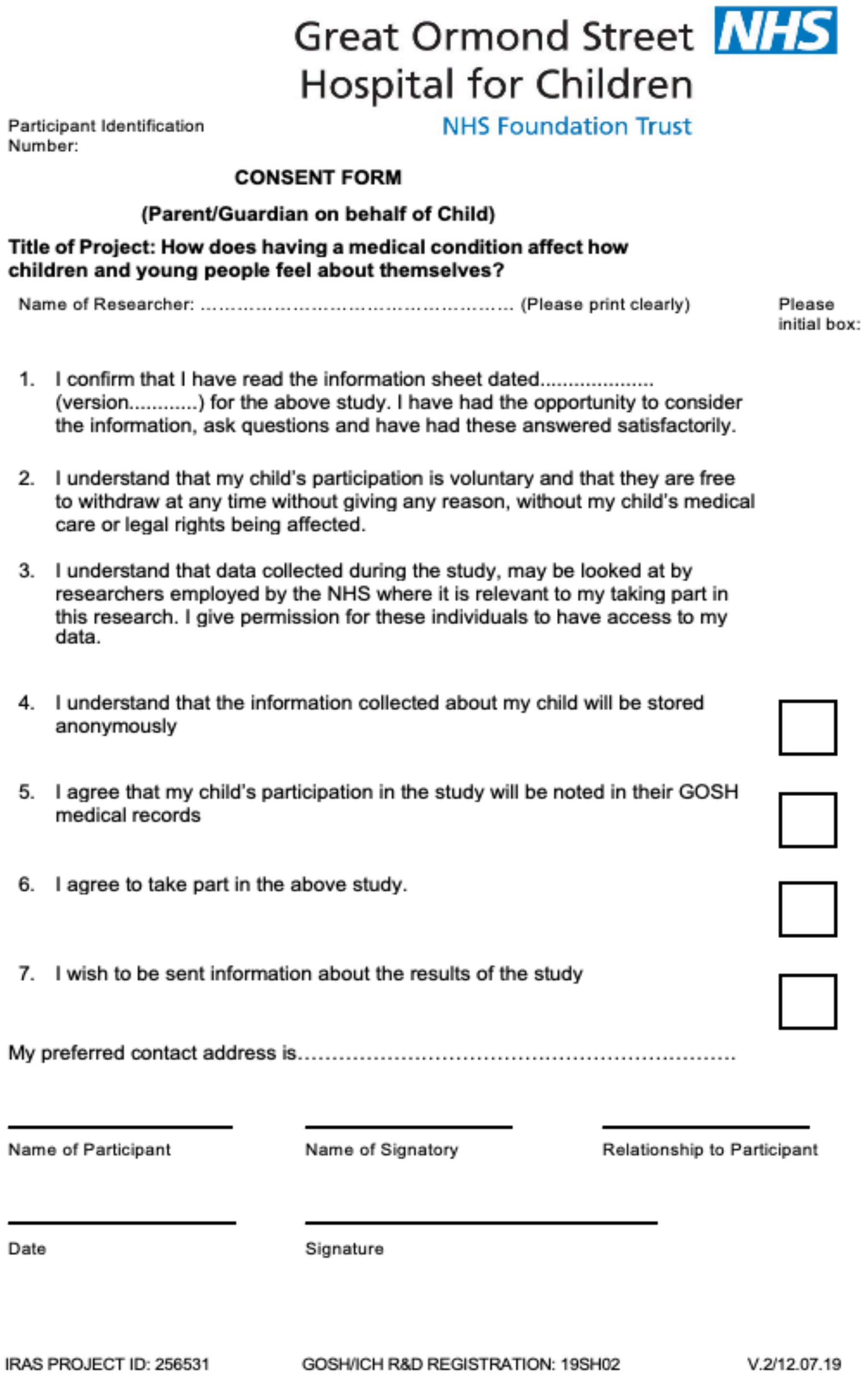
How will taking part in this study help others?
Helping us to understand how it feels to have a medical condition will help us to look after the feelings of all patients at GOSH who might feel the same way. We hope that this study will improve how we look after patients in GOSH so that they can feel 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: [redacted] at GOSH, on [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 O: Parent Consent Form



Great Ormond Street Hospital for Children
NHS
NHS Foundation Trust

Participant Identification Number:

CONSENT FORM

(Parent/Guardian on behalf of Child)

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

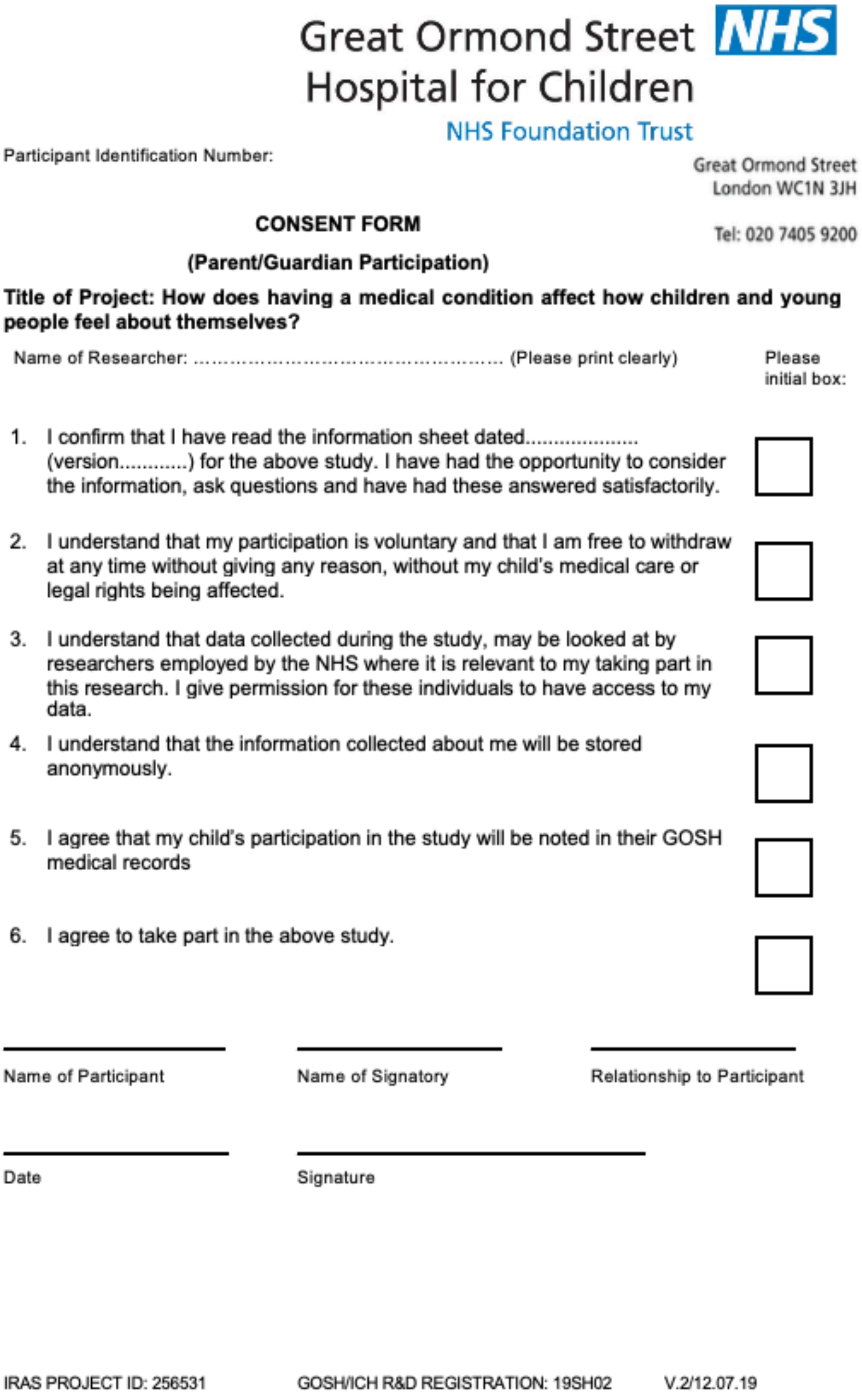
Relationship to Participant

Date

Signature

IRAS PROJECT ID: 256531 GOSH/ICH R&D REGISTRATION: 19SH02 V.2/12.07.19

Appendix O (cont.): Parent Consent Form



Great Ormond Street NHS
Hospital for Children
NHS Foundation Trust

Great Ormond Street
London WC1N 3JH

Tel: 020 7405 9200

Participant Identification Number:

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:

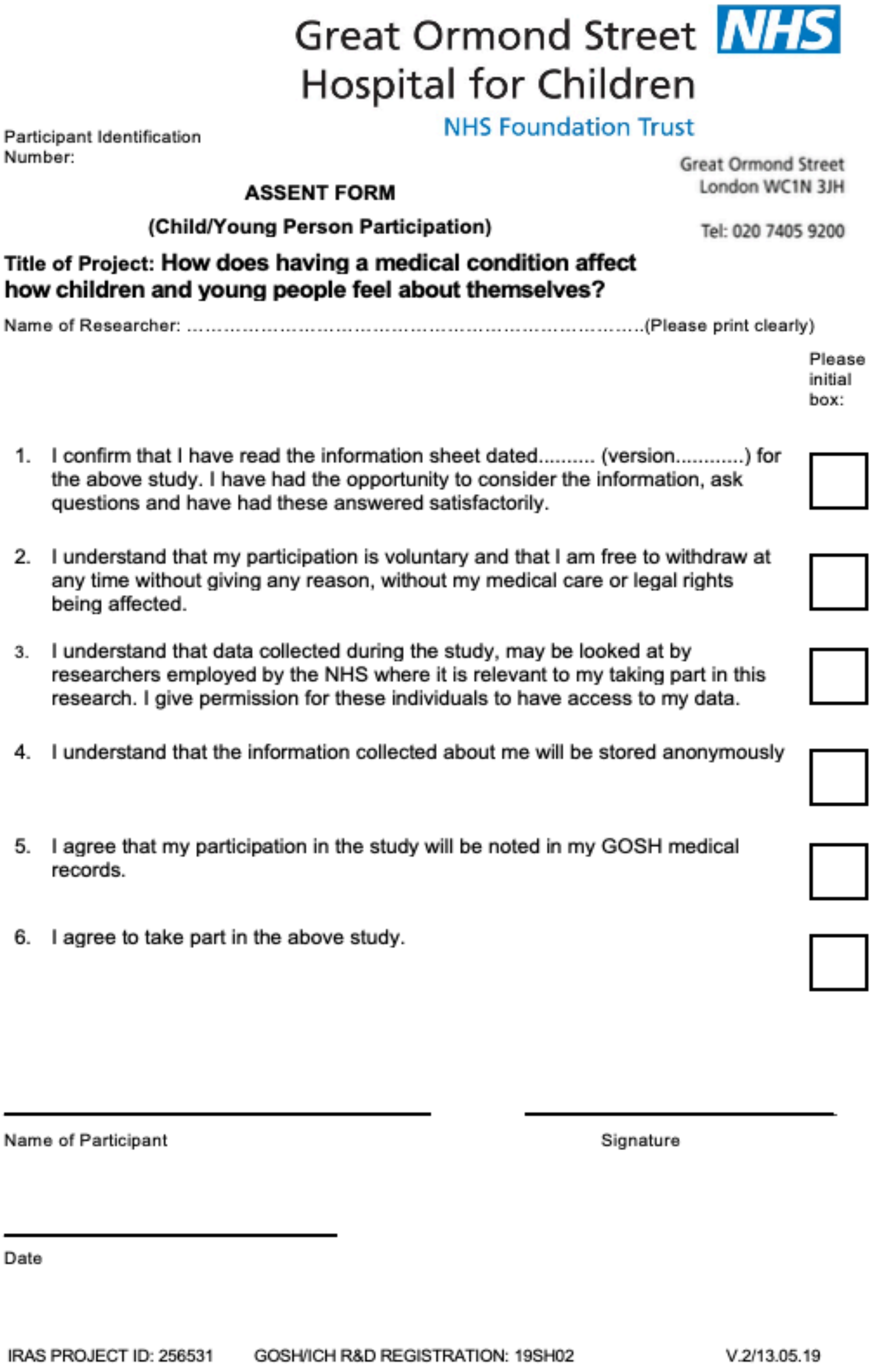
1. I confirm that I have read the information sheet dated.................... (version............) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. ☐
2. I understand that my participation is voluntary and that I am 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 ________

IRAS PROJECT ID: 256531 GOSH/ICH R&D REGISTRATION: 19SH02 V.2/12.07.19

Appendix P: Child and Young Person Consent Form



Great Ormond Street Hospital for Children NHS

NHS Foundation Trust

Great Ormond Street
London WC1N 3JH

Tel: 020 7405 9200

Participant Identification Number:

ASSENT FORM

(Child/Young Person 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 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 ______________________

IRAS PROJECT ID: 256531 GOSH/ICH R&D REGISTRATION: 19SH02 V.2/13.05.19

Appendix Q: Kolmogorov Tests of Normality

Tests of Normality

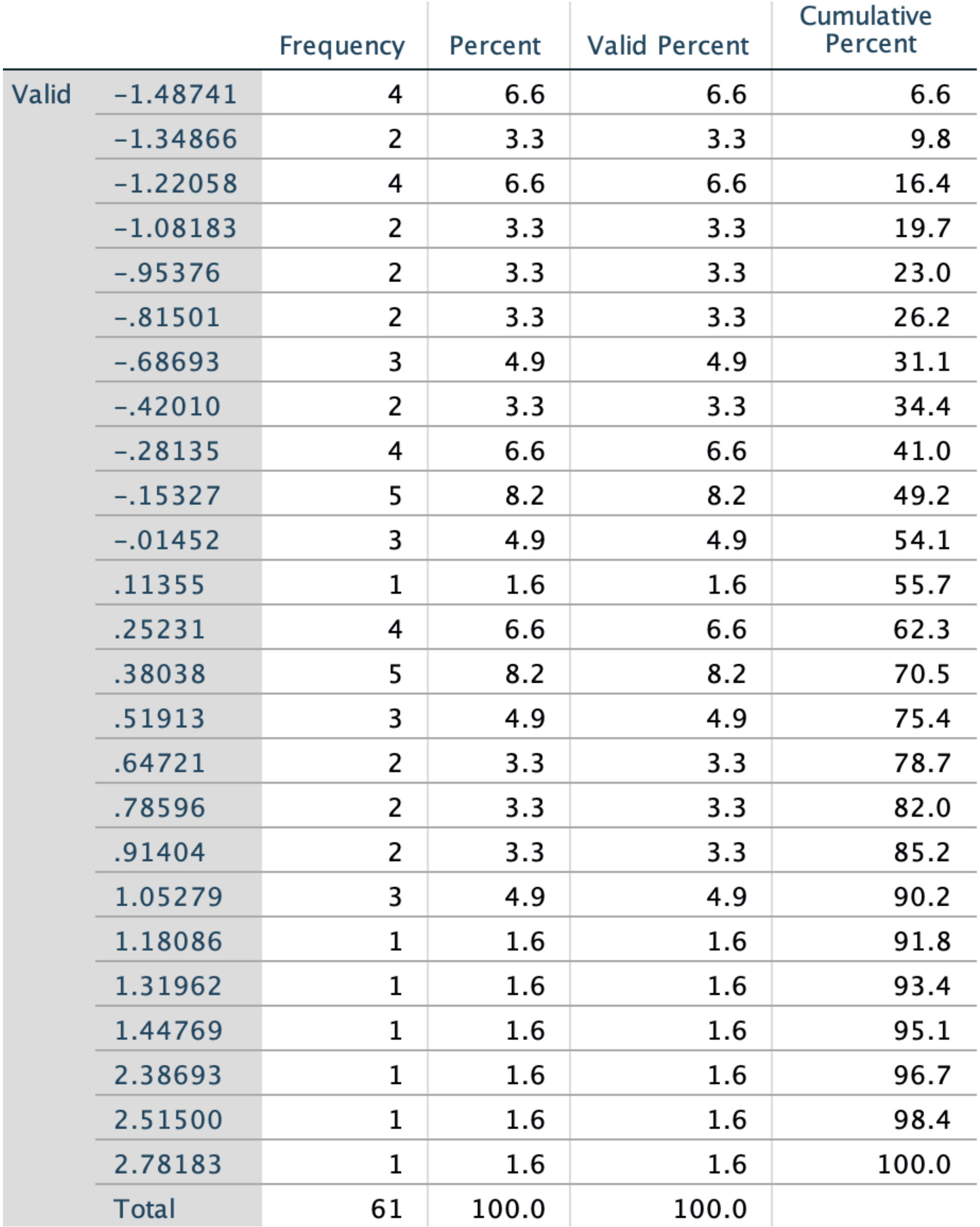
	Kolmogorov-Smirnov[a]		
	Statistic	df	Sig.
SelfStigmaScore	.099	61	.200*
ConcealmentScore	.068	61	.200*
Illness Attitudes	.083	61	.200*
EmotionalPeerDifficulties	.152	61	.001

*. This is a lower bound of true significance

[a] = Lilliefors Significance Correction

Appendix R: Assessing for Outliers

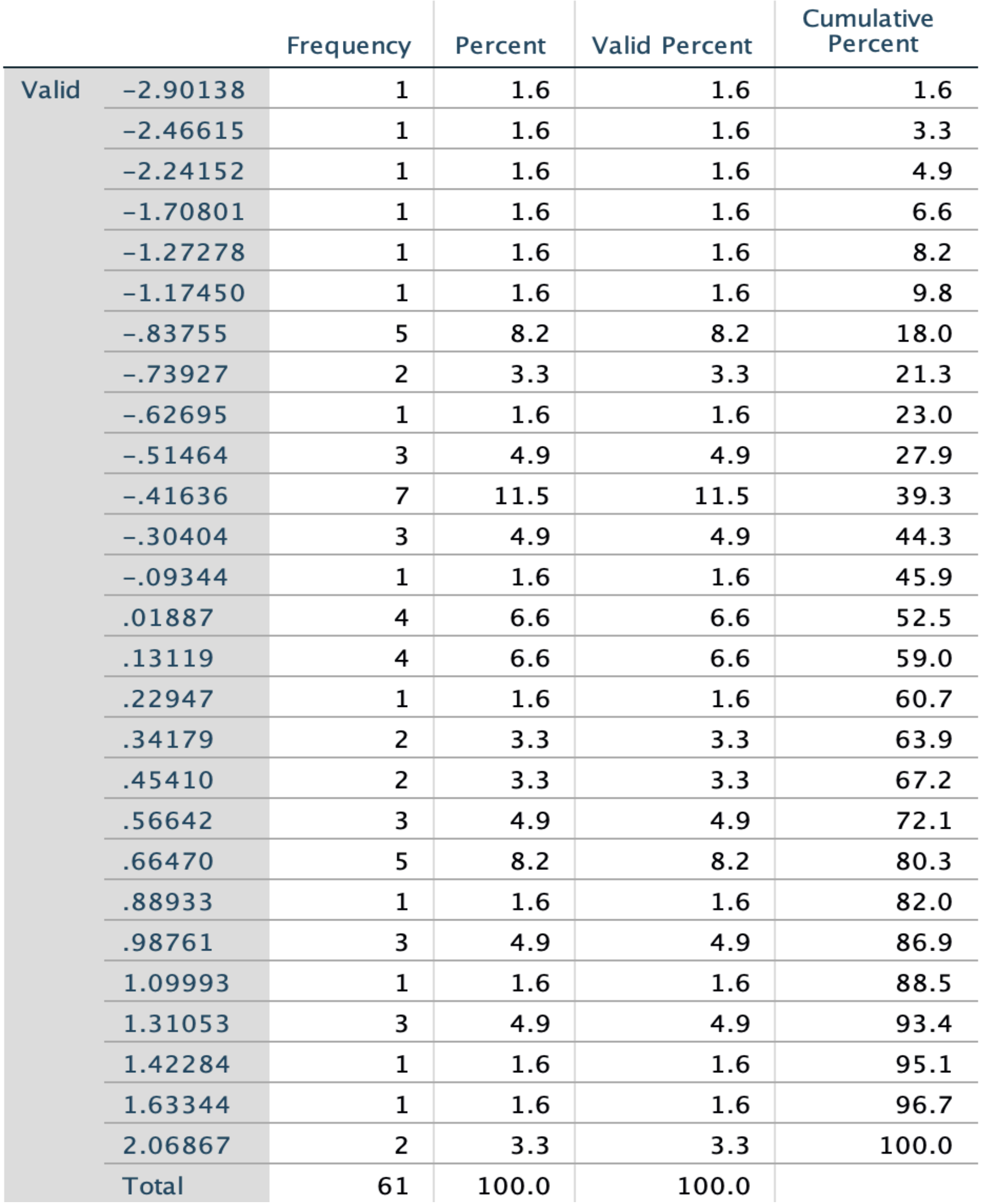
Zscore: Self-Stigma Scale



		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	-1.48741	4	6.6	6.6	6.6
	-1.34866	2	3.3	3.3	9.8
	-1.22058	4	6.6	6.6	16.4
	-1.08183	2	3.3	3.3	19.7
	-.95376	2	3.3	3.3	23.0
	-.81501	2	3.3	3.3	26.2
	-.68693	3	4.9	4.9	31.1
	-.42010	2	3.3	3.3	34.4
	-.28135	4	6.6	6.6	41.0
	-.15327	5	8.2	8.2	49.2
	-.01452	3	4.9	4.9	54.1
	.11355	1	1.6	1.6	55.7
	.25231	4	6.6	6.6	62.3
	.38038	5	8.2	8.2	70.5
	.51913	3	4.9	4.9	75.4
	.64721	2	3.3	3.3	78.7
	.78596	2	3.3	3.3	82.0
	.91404	2	3.3	3.3	85.2
	1.05279	3	4.9	4.9	90.2
	1.18086	1	1.6	1.6	91.8
	1.31962	1	1.6	1.6	93.4
	1.44769	1	1.6	1.6	95.1
	2.38693	1	1.6	1.6	96.7
	2.51500	1	1.6	1.6	98.4
	2.78183	1	1.6	1.6	100.0
	Total	61	100.0	100.0	

Appendix R (cont.): Assessing for Outliers

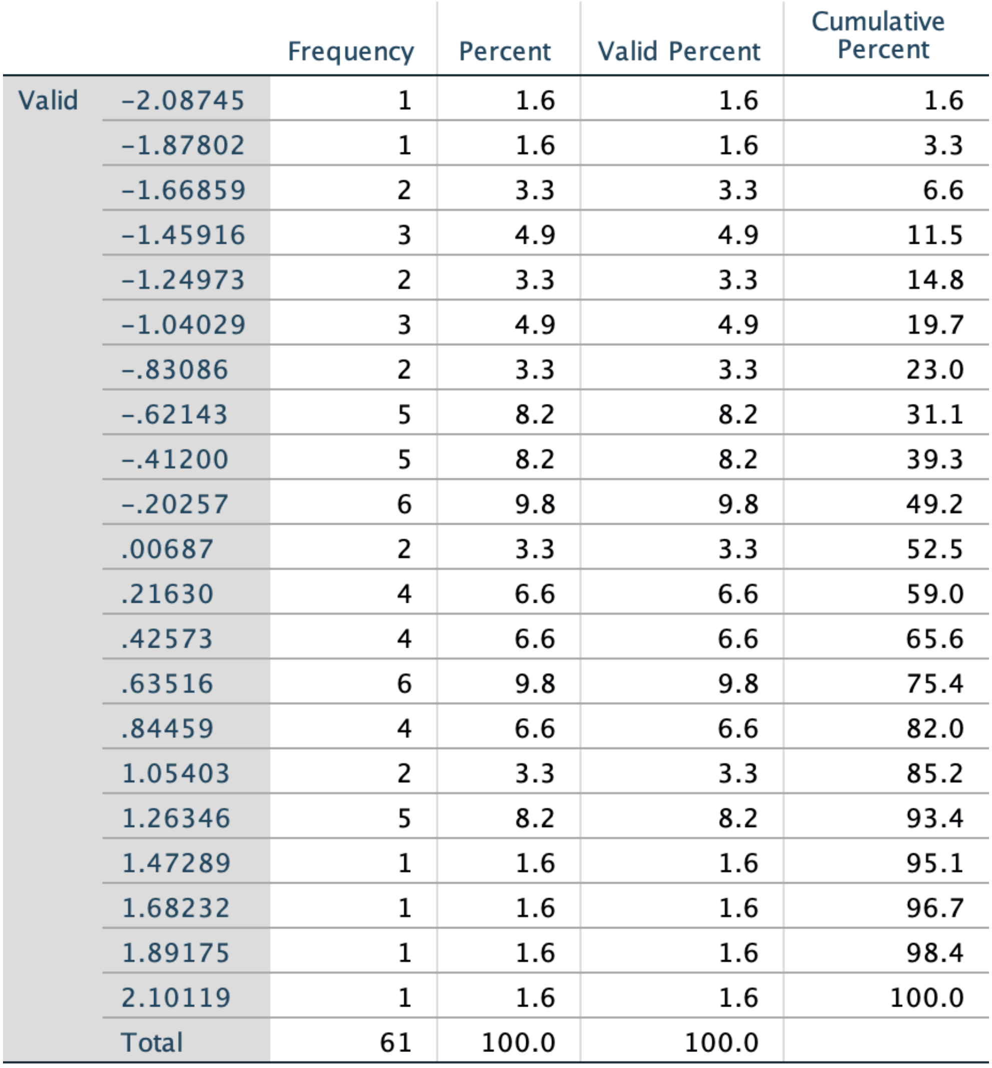
Zscore: Illness Attitudes



		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	-2.90138	1	1.6	1.6	1.6
	-2.46615	1	1.6	1.6	3.3
	-2.24152	1	1.6	1.6	4.9
	-1.70801	1	1.6	1.6	6.6
	-1.27278	1	1.6	1.6	8.2
	-1.17450	1	1.6	1.6	9.8
	-.83755	5	8.2	8.2	18.0
	-.73927	2	3.3	3.3	21.3
	-.62695	1	1.6	1.6	23.0
	-.51464	3	4.9	4.9	27.9
	-.41636	7	11.5	11.5	39.3
	-.30404	3	4.9	4.9	44.3
	-.09344	1	1.6	1.6	45.9
	.01887	4	6.6	6.6	52.5
	.13119	4	6.6	6.6	59.0
	.22947	1	1.6	1.6	60.7
	.34179	2	3.3	3.3	63.9
	.45410	2	3.3	3.3	67.2
	.56642	3	4.9	4.9	72.1
	.66470	5	8.2	8.2	80.3
	.88933	1	1.6	1.6	82.0
	.98761	3	4.9	4.9	86.9
	1.09993	1	1.6	1.6	88.5
	1.31053	3	4.9	4.9	93.4
	1.42284	1	1.6	1.6	95.1
	1.63344	1	1.6	1.6	96.7
	2.06867	2	3.3	3.3	100.0
	Total	61	100.0	100.0	

Appendix R (cont.): Assessing for Outliers

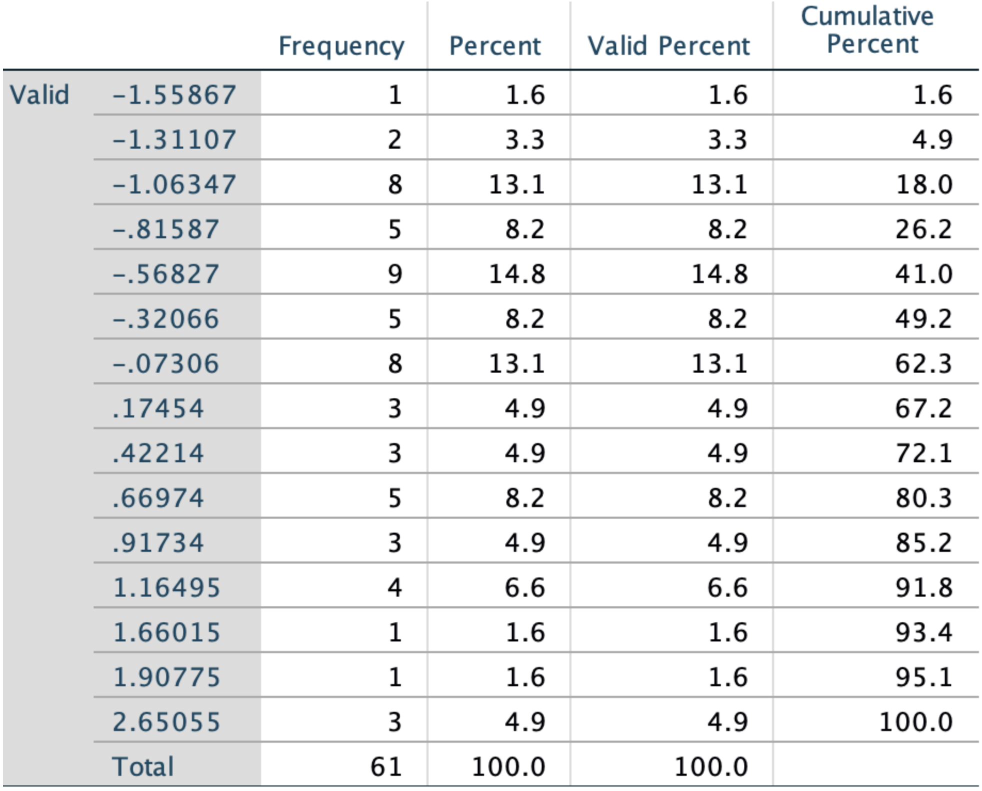
Zscore: Concealment Score



		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	-2.08745	1	1.6	1.6	1.6
	-1.87802	1	1.6	1.6	3.3
	-1.66859	2	3.3	3.3	6.6
	-1.45916	3	4.9	4.9	11.5
	-1.24973	2	3.3	3.3	14.8
	-1.04029	3	4.9	4.9	19.7
	-.83086	2	3.3	3.3	23.0
	-.62143	5	8.2	8.2	31.1
	-.41200	5	8.2	8.2	39.3
	-.20257	6	9.8	9.8	49.2
	.00687	2	3.3	3.3	52.5
	.21630	4	6.6	6.6	59.0
	.42573	4	6.6	6.6	65.6
	.63516	6	9.8	9.8	75.4
	.84459	4	6.6	6.6	82.0
	1.05403	2	3.3	3.3	85.2
	1.26346	5	8.2	8.2	93.4
	1.47289	1	1.6	1.6	95.1
	1.68232	1	1.6	1.6	96.7
	1.89175	1	1.6	1.6	98.4
	2.10119	1	1.6	1.6	100.0
	Total	61	100.0	100.0	

Appendix R (cont.): Assessing for Outliers

Zscore 1: Emotional and Peer Difficulties



		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	-1.55867	1	1.6	1.6	1.6
	-1.31107	2	3.3	3.3	4.9
	-1.06347	8	13.1	13.1	18.0
	-.81587	5	8.2	8.2	26.2
	-.56827	9	14.8	14.8	41.0
	-.32066	5	8.2	8.2	49.2
	-.07306	8	13.1	13.1	62.3
	.17454	3	4.9	4.9	67.2
	.42214	3	4.9	4.9	72.1
	.66974	5	8.2	8.2	80.3
	.91734	3	4.9	4.9	85.2
	1.16495	4	6.6	6.6	91.8
	1.66015	1	1.6	1.6	93.4
	1.90775	1	1.6	1.6	95.1
	2.65055	3	4.9	4.9	100.0
	Total	61	100.0	100.0	

Appendix S: Demographic and Medical Outputs

T-Tests

Self-Stigma					
Demographic Variable	*n*	M	SD	t	p
Gender				1.52	.14
Female	*31*	2.57	.94		
Male	*30*	2.21	.92		
Clinic Type				-2.00	.05
Dermatology	*34*	2.59	.92		
Urology	*27*	2.12	.91		
Ethnicity				.32	.75
White	*46*	2.42	1.01		
Black, Asian or other ethnicity	*15*	2.33	.68		
Other Medical Condition				.47	.64
Yes	*18*	2.48	.91		
No	*43*	2.36	.47		
Data Collection				1.01	.32
Face to Face	*49*	2.45	.98		
Remote	*12*	2.15	.73		

Concealment					
Demographic Variable	*n*	M	SD	t	p
Gender				.74	.46
Female	*31*	2.49	.65		
Male	*30*	2.36	.72		
Clinic Type				-.54	.59
Dermatology	*34*	2.47	.71		
Urology	*27*	2.37	.65		
Ethnicity				-.73	.47
White	*46*	2.40	.74		
Black, Asian or other ethnicity	*15*	2.51	.48		
Other Medical Condition				.39	.70
Yes	*18*	2.48	.69		
No	*43*	2.40	.69		
Data Collection				.99	.33
Face to Face	*49*	2.47	.69		
Remote	*12*	2.24	.67		

Illness Attitudes					
Demographic Variable	*n*	M	SD	t	p
Gender				-2.29	.03*
Female	*31*	3.02	.67		

Male	*30*	3.43	.70		
Clinic Type				3.50	.00*
Dermatology	*35*	2.95	.58		
Urology	*26*	3.54	.73		
Ethnicity				-.60	.55
White	*46*	3.18	.73		
Black, Asian or Other Ethnicity	*15*	3.31	.65		
Other Medical Condition				-.93	.35
Yes	*18*	3.08	.75		
No	*43*	3.27	.70		
Data Collection				-.64	.53
Face to face	*49*	3.19	.72		
Remote	*12*	3.33	.68		

Mann Whitney U

Emotional and Peer Difficulties

Demographic Variable	*n*	Mdn	U	z	p
Gender			411.5	-.78	.44
Female	*31*	5			
Male	*30*	6			
Clinic Type			471.	.17	.86
Dermatology	*34*	5.5			
Urology	*27*	6			
Ethnicity			323.	-.37	.71
White	*46*	6			
Black, Asian or Other ethnicity	*15*	5			
Other Medical Condition			255.	-2.1	.04*
Yes	*18*	8			
No	*43*	5			
Data Collection			255.	-.71	.48
Face to face	*49*	6			
Remote	*12*	4.5			

Appendix S (cont.): Demographic and Medical Output

ANOVAs

Self-Stigma

Demographic Variable	*n*	M	SD	F	p
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				2.27	.07
Eczema	*19*	2.50	.90		
Epidermolysis Bullosa	*9*	2.37	.81		
Psoriasis	*6*	3.31	1.02		
Bladder Diagnosis	*12*	2.20	.69		
Kidney Diagnosis	*15*	2.06	1.04		

Concealment

Demographic Variable	*n*	M	SD	F	p
School Year				0.93	.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*	2.72	.57		
Kidney Diagnosis	*15*	2.10	.58		

Illness Attitudes

Demographic Variable	*n*	M	SD	F	p
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	1.17		
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				3.71	.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	.79		

Appendix S (cont.): Demographic and Medical Output

Kruskal-Wallis Test

Emotional and Peer problems				
Demographic Variable	*n*	Mdn	X^2	p
School Year			6.78	.08
Year 3-4	*7*	5		
Year 5-6	*16*	4		
Year 7-8	*18*	6.5		
Year 9-10	*20*	6		
Time of Onset			4.04	.13
Present at Birth	*24*	4		
Less than 5 years old	*23*	6		
5 years or older	*14*	7.5		
Hospital Attendance			8.49	.01*
1-2 times per year	30	4		
3-10 times per year	25	7		
More than 10 times per year	6	8.5		
Medical Condition			3.01	.56
Eczema	*19*	6		
Epidermolysis Bullosa	*9*	4		
Psoriasis	*6*	5.5		
Bladder Diagnosis	*12*	5.5		
Kidney Diagnosis	*15*	6		