Communication in Sickle Cell Disease: A Meta-Synthesis of Child Perspectives and a Qualitative Exploration of Parent Experience

A thesis submitted to The University of Manchester for the degree of Doctorate in Clinical Psychology (ClinPsyD) in the Faculty of Biology, Medicine and Health

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Thesis Abstract

Communication in Sickle Cell Disease: A Meta-Synthesis of Child Perspectives and a Qualitative Exploration of Parent Experience

Joanne Middleton

Doctorate in Clinical Psychology (ClinPsyD) The University of Manchester

October 2016

This thesis explores communication with children affected by sickle cell disease about their condition from the perspectives of both children and parents. It includes three papers: A literature review, an empirical paper and a critical appraisal. Papers one and two have been prepared for submission to Social Science and Medicine and Qualitative Health Research, respectively. Paper one is a meta-synthesis of qualitative literature investigating experiences of communication from the perspective of children with sickle cell disease. A systematic literature search revealed nine relevant papers, which were synthesised by extracting findings related to communication about sickle cell disease. Children were found to receive inconsistent messages about their condition from different personal and professional groups. Communication about the prognosis of sickle cell disease and the social acceptability of the condition differed across the groups. The implications for children’s understandings of their condition and their adjustment are discussed. Paper two presents an empirical study of parental communication experiences with children affected by sickle cell disease. Twelve interviews were conducted and subject to inductive thematic analysis which was applied within a contextualist epistemological framework. Parents described skills in ‘coaching’ their child to negotiate the various challenges associated with managing sickle cell disease. They also described ways in which they avoided challenging topics of communication such as inheritance, the risk of comorbid disease and the life-long nature of the condition. The findings suggest a need for healthcare professionals to support parents in overcoming barriers to talking about difficult topics. This may facilitate more consistent communication between parents and professionals, which has implications for improving child wellbeing and adjustment. Paper three is a reflective piece and is not intended for publication. It critically evaluates papers one and two and discusses the joint implications of the findings for research and clinical practice. Reflections on the experience of conducting a meta-synthesis and an empirical qualitative study are offered in the context of personal and professional development.
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No portion of the work referred to in the thesis has been submitted in support of an application for another degree or qualification of this or any other university or other institute of learning
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Acknowledgements

A number of people have provided invaluable support in various ways to make this thesis possible. I am hugely grateful to Fiona Ulph for the time she has given in supporting this project. Fiona, your wealth of experience and knowledge about qualitative research and genetic conditions has been invaluable as has your genuine interest and enthusiasm for my work. Thanks also to Rachel Calam for your encouragement and guidance from start to finish. Thank you to Sonia Patel for your support of the project overall, but particularly for your advice and assistance during recruitment. I would like to thank each of the twelve parents who participated in the study, many of whom welcomed me warmly into their homes and courageously shared their stories.

Returning to the ClinPsyD from maternity leave has been a huge challenge that I have managed thanks to my wonderfully supportive colleagues and friends, Annukka, Katie, Miriam and Sarah. I am so proud of us all and thankful to have been able to support one another during the past year.

I am very lucky to have a supportive family and wonderful parents who have given me so many opportunities and have supported all my endeavours - great and small. Thank you for always being there, both in person or at the end of the phone, and for reminding me to believe in myself. Rob, you’ve been alongside me since the start of this rather bumpy journey and have witnessed the blood, sweat and tears along the way. Thank you for easing my troubles and keeping me grounded in what matters the most – the health and happiness of our wonderful little family. You and Thomas have been the light at the end of every day and this thesis is dedicated to you both. My time is ours, once again.
Child perspectives of communication in sickle cell disease: A meta-synthesis of qualitative research

Paper 1 has been prepared for submission to Social Science and Medicine in accordance with the journal guidelines for contributors (Appendix 1).

**Word Count:** 6108 including all tables & references
Abstract

Sickle Cell Disease (SCD) is a genetic condition with symptoms of pain and fatigue that begin in early childhood. The disease presents a number of communication challenges for parents, from explaining the physiology of the condition to informing the child of the impact it is likely to have on their future. Few studies have explored child communication in SCD specifically and instead tend to include SCD within wider samples of genetic conditions. This review is the first to synthesise the experiences of communication with children about SCD specifically, by extracting findings related to SCD from both general and specific studies. The aim was to take the perspective of the child and to consider how they experience communication. A systematic literature search of seven databases was conducted using predefined search terms. Nine papers were synthesised using an interpretive approach based on Noblit and Hare’s (1988) meta-ethnographic principles. The synthesis focussed on the similarities and differences between studies, and thus between the different groups of people communicating with the child. A refutational analysis identified four common themes: Children’s experience of ‘informing and being informed’ about SCD, children’s ‘preferences’ in the communication they receive, the ‘functions’ of others’ communication and finally, the ‘consequences’ of this communication on the child’s adjustment. Children were found to receive ‘mixed messages’ about their condition from different groups of people (i.e. parents, school teachers, peers and healthcare professionals). Mixed messages were related to the severity of SCD and the acceptability of the condition within society. Implications for a child’s understanding and adjustment to their condition are discussed with suggestions for improving healthcare practice.

Abstract Word Count: 263
Research highlights:

- Children with SCD receive inconsistent messages from different personal and professional groups.
- Inconsistencies in information and communication may impact on their identity and adjustment.
- Parents were found to have avoided communication about some topics.
- Adults supporting children may need help to facilitate open and consistent communication.
- Interventions aimed at addressing inconsistencies offer to help children better adapt to SCD.

Key words: Sickle Cell Disease, Child, Communication, Qualitative, Meta-synthesis, Meta-ethnography
Introduction

Sickle cell disease (SCD) is a genetic blood disorder, most commonly affecting people of African and African Caribbean origin, although cases also occur in families originating from the Middle East, India and the Eastern Mediterranean (De, 2005). SCD is the most common genetic condition in England, affecting more than 1 in 2,000 children; however, the birth prevalence in some urban areas may be as high as 1 in 300 (NHS Sickle Cell and Thalassaemia Screening Programme, 2010). Symptoms include acute episodes of pain, known as ‘crises’ which are caused by small blood vessel blockages and can, over time, result in irreversible organ damage and associated dysfunction (Franklin, 1990; Powars, Chan, Hiti, Ramicone & Johnson, 2005). Children affected by SCD are subject to numerous investigations, medications and procedures to manage their symptoms and prevent crises. Like many other chronic conditions, SCD requires careful monitoring by medical teams and by the family at home (Herzer, Godiwala, Hommel et al., 2010). Children also rely on their school’s understanding of their condition and their increased absence rates, which have academic and social implications (Atkin & Ahmad, 2001; Lightfoot, Wright & Slopper, 1999).

As an incurable illness affecting minority ethnic groups, disability and racism are common sources of discrimination described by those affected (Bediako & Moffitt, 2011). Illness related stigma about SCD has been found within the black and minority ethnic (BME) community due to historic stereotypes of people with SCD living short, limited lives and therefore lacking reproductive appeal (Burnes, Antle, Williams & Cook, 2008; Jenerette, Funk & Murdaugh, 2005; Thomas & Taylor, 2002; Ulph, Cullinan, Qureshi & Kai, 2011). Affected children and families are therefore faced with the challenge of integrating both the physical and socio-cultural aspects of the
condition into their lives. Illness related conversations offer families the opportunity to enhance a child’s understanding about their condition and dispel any myths, thereby promoting better adjustment (Rushforth, 1999). Research in other genetic conditions has demonstrated that children, who are not included in discussions about genetic risk, may be affected by low self-esteem (Fanos, Davis & Puck, 2001; McConkie-Rosell, Heise & Spiridigliozi, 2009) and poor family cohesion (Sobell & Cowan, 2000; 2003). Furthermore, the complexity of disclosing genetic risk information can result in delayed or non-disclosure about genetic conditions leading to misunderstanding, anxiety and increased family tensions (Rowland & Metcalfe, 2013; Ulph et al., 2011).

Communication about genetic risk has been studied more extensively in parents of sickle cell carriers who have been found to hold positive attitudes about open communication, but appeal for more support from healthcare professionals in the process (Ulph et al., 2011). Healthcare professionals have however faced difficulties in supporting open family communication due to parental efforts to shield children from potentially upsetting information (Ulph, Leong, Glazebrook & Townsend, 2010).

As they become socialised to their condition, children are likely to experience various styles of communication from different points of contact. For example, a healthcare professional’s role is to hold open and accurate discussions with families and children; however, parents may be withholding information in order to protect their child from unwanted distress (Ayme, Macquart- Moulin, Julian-Reynier, Chabal & Giraud, 1993, Ulph et al., 2010). It is important to consider that these interactions and communication styles may be happening alongside each other, carrying the risk of children receiving conflicting information. This is an issue that has not been addressed previously and forms the purpose of this paper.
Aims

By employing techniques of meta-ethnography (Noblit & Hare, 1988), this review aimed to understand communication from the perspective of the affected child with SCD. The aim was to integrate the findings from individual studies in order to understand communication more holistically, taking the perspective of the affected child who is likely to be communicating about SCD with a number of groups including family, peers and healthcare professionals. A number of specific research questions were developed to guide the exploration:

- Who does the child with SCD communicate with about their condition?
- How do different groups of people communicate with the child?
- In what ways does this communication differ?
- What is the child’s experience of these multiple interactions?
- What implications can be made about the child’s adjustment based on these findings?

Method

A number of methodologies have been suggested for synthesising qualitative research (Downe, 2008). This paper presents a meta-synthesis that employed principles of meta-ethnography (Box 1; Noblit & Hare, 1988) and Schutz’s (1962) notions of first and second order constructs. This approach to synthesising the literature was chosen based on its ability to produce new and integrative interpretations of existing findings that are more substantial than those resulting from individual investigations (Finfgeld, 2003).
Having decided upon the research questions (Step 1), inclusion and exclusion criteria (Box 2) were agreed upon by the authors and the literature was systematically searched (Step 2). Search terms were developed using the SPIDER tool (Cooke, Smith & Booth, 2012), which provides a framework for choosing search terms in qualitative research based on the sample, phenomenon of interest, design, evaluation outcomes and research type. A flow diagram of the search process is illustrated in Figure 1.

<table>
<thead>
<tr>
<th>Box 1. Noblit and Hare’s analytic strategy for meta-ethnography</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Getting started</td>
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<tr>
<td>2. Deciding what is relevant to the initial interest</td>
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<tr>
<td>3. Reading the studies</td>
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<tr>
<td>4. Determining how the studies are related</td>
</tr>
<tr>
<td>5. Translating the studies into each other</td>
</tr>
<tr>
<td>a. Reciprocal translation</td>
</tr>
<tr>
<td>b. Refutational translation</td>
</tr>
<tr>
<td>6. Synthesising translations into a ‘line of argument’</td>
</tr>
<tr>
<td>7. Expressing the synthesis</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Box 2. Inclusion and Exclusion Criteria</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Inclusion criteria</strong></td>
</tr>
<tr>
<td>Original peer reviewed research articles</td>
</tr>
<tr>
<td>Studies of communication within families with at least one child affected by SCD</td>
</tr>
<tr>
<td>Studies of communication within families about genetic risk for SCD</td>
</tr>
<tr>
<td>Studies of communication between healthcare professionals, school teachers, peers and affected children about SCD or genetic risk</td>
</tr>
<tr>
<td>Communication with children under age 18</td>
</tr>
<tr>
<td><strong>Exclusion criteria</strong></td>
</tr>
<tr>
<td>Literature reviews, meta analyses or metasyntheses</td>
</tr>
<tr>
<td>Quantitative studies</td>
</tr>
<tr>
<td>Studies that lack clarity on findings that are specific to SCD</td>
</tr>
<tr>
<td>Studies of communication with children aged 19 and over</td>
</tr>
<tr>
<td>Studies of carriers or siblings</td>
</tr>
</tbody>
</table>
Figure 1. Systematic literature search flow diagram

Search Terms:
Word truncations: famil*, parent, child*, sibling, adolescent, healthcare professional, peer communicat*, discuss*, inform*, disclos*, sickle cell*, hemoglobinopath*, ‘genetic condition’
NOT: cancer, cystic fibrosis, autism*, learning disability

Databases Searched
EBSCOhost databases (CINAHL, ERIC), Ovid Databases (Embase, Medline, Psychinfo), Web of Science, British Nursing Index, Free hand searches.

Not original peer reviewed research articles
N = 3

Quantitative Studies
N = 3

Not focussed on family communication in SCD
N = 16

Studies included in the analysis
N = 9
Search terms were developed based on truncations of words related to communication, children, families, school, peers, sickle cell disease and genetic conditions. Wild card and Boolean Search Operators were used to capture the most relevant studies. A total of seven databases were searched, which included OVID databases (MEDLINE, EMBASE and PsychINFO), EBSCOhost databases (CINAHL and ERIC), Web of Science and the British Nursing Index (BNI). Free hand searches of reference lists were completed in addition to systematic database searching to maximise inclusion. Searches were run in September 2015 and updated in August 2016. Publications from 2005 to present were searched for to ensure findings were applicable to current understandings and perspectives on SCD. Other limits such as ‘English language’, ‘peer reviewed journals’ and ‘qualitative research’ were applied to ensure the search results included evidence based research that was relevant to the research questions.

Initial searches revealed 475 papers. Following the removal of duplicates [n = 82] the titles and abstracts of the remaining 392 studies were screened for relevance. This process resulted in 37 potentially relevant articles, all of which were downloaded electronically in full text and considered according to inclusion and exclusion criteria (Box 2). The nine studies meeting inclusion criteria were subject to quality appraisal, which is described in full below. The aim of this stage was to consider the quality of the studies included and not to exclude any that obtained a poor quality rating.

**Quality and characteristics of papers**

‘Relevance and Transferability’. The complete quality analysis is shown in Appendix 2, within which rating of quality using a three-star system led to the classification of studies according to categories A, B, C or D (as used by Downe, 2008). Quality ratings are shown alongside the key features of each study in Table 1. All nine papers were agreed to warrant inclusion in the meta-synthesis, based on their theoretical clarity and methodological rigour. The synthesis includes seven samples from nine studies. Two of the papers, Gallo, Angst, Knafl, Hadley and Smith (2005) and Gallo, Angst and Knafl (2009), published findings on a sample that included the same 29 parents of children with sickle cell disease. Three of the papers, Metcalfe, Plumridge, Coad, Shanks & Gill (2011), Plumridge, Metcalfe, Coad & Gill (2011a) and Plumridge, Metcalfe, Coad and Gill (2011b) published findings on a sample that included 33 families of children affected by SCD. Although this may place some limitations on the review, it was agreed to include all of the studies to maximise inclusivity and allow the distinct findings of each paper to be synthesised in their own right.
Table 1. Characteristics and quality ratings of included studies

<table>
<thead>
<tr>
<th>Authors</th>
<th>Title</th>
<th>Method/analysis</th>
<th>Country</th>
<th>Participants</th>
<th>Conditions included</th>
<th>Quality Rating</th>
</tr>
</thead>
<tbody>
<tr>
<td>Gallo et al., (2005)</td>
<td>Parents sharing information with their children about genetic conditions</td>
<td>Semi-structured interviews</td>
<td>USA</td>
<td>Parents of children with various genetic conditions (N = 139)</td>
<td>SCD (29), PKU (16), CF (15), NF (11), Hemophilia (6), Thalassemia (4), Marfan Syndrome (4), Von Willebrand disease (1)</td>
<td>B</td>
</tr>
<tr>
<td>Gallo et al., (2009)</td>
<td>Information management in families who have a child with a genetic condition</td>
<td>Mixed methods: Semi structured interviews &amp; structured measures of family functioning</td>
<td>USA</td>
<td>Parents of children with various genetic conditions (N = 142)</td>
<td>SCD (29), PKU (16), CF (16), NF (10), Bleeding disorders (7), Thalassemia (4), Marfan Syndrome (4).</td>
<td>A</td>
</tr>
<tr>
<td>Gallo et al. (2010)</td>
<td>Healthcare professionals’ views of sharing information with families who have a child with a genetic condition</td>
<td>Semi-structured interviews: Thematic analysis</td>
<td>USA</td>
<td>37 healthcare professionals of children with genetic conditions (including SCD)</td>
<td>As above. Participants were caregivers of the above sample</td>
<td>A</td>
</tr>
<tr>
<td>Study</td>
<td>Title</td>
<td>Methodology</td>
<td>Country</td>
<td>Sample Description</td>
<td>Findings</td>
<td></td>
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<td>-----------------------------------------------------------------------</td>
<td>------------------------------------</td>
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<td>------------------------------------------------------------------------------------</td>
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<td></td>
</tr>
<tr>
<td>Graff et al., (2012)</td>
<td>Exploring family communication about SCD in adolescence</td>
<td>Focus Group</td>
<td>USA</td>
<td>Parents (12), Affected adolescents (N= 11) &amp; Siblings (N = 11)</td>
<td>SCD</td>
<td></td>
</tr>
<tr>
<td>Metcalfe et al., (2011)</td>
<td>Parents’ and children’s communication about genetic risk: a qualitative study, learning from families’ experiences.</td>
<td>Semi Structured interviews</td>
<td>UK</td>
<td>Families (N = 33) including parents (N = 52) and children (N = 33) affected or at risk from genetic conditions</td>
<td>NF (4), HD (7), *HbO 1(6), FAP (6), DMD (6), CF (4)</td>
<td></td>
</tr>
<tr>
<td>Plumridge et al., (2011)</td>
<td>The role of support groups in facilitating families in coping with a genetic condition and in discussion of genetic risk information</td>
<td>Semi structured interviews</td>
<td>UK</td>
<td>Families (N = 33) including parents (N = 52) and children (N = 33) affected or at risk from genetic conditions</td>
<td>NF (4), HD (7), *HbO (6), FAP (6), DMD (6), CF (4)</td>
<td></td>
</tr>
<tr>
<td>Atkin &amp; Ahmad (2001)</td>
<td>Living a ‘normal’ life: young people with thalassemia major or SCD</td>
<td>Semi structured interviews</td>
<td>UK</td>
<td>26 interviews (18 SCD)</td>
<td>SCD (18) and Thalassaemia major (8)</td>
<td></td>
</tr>
<tr>
<td>Dyson et al., (2010)</td>
<td>Disclosure and sickle cell disorder: A mixed methods study of the young person with sickle cell at school</td>
<td>569 questionnaires/ 40 interviews</td>
<td>UK</td>
<td>40 Young people aged 11 to 25 with SCD</td>
<td>SCD</td>
<td></td>
</tr>
<tr>
<td>Jacob et al., (2012)</td>
<td>Facilitating paediatric patient provider communications using wireless technology in children and adolescents with sickle cell disease</td>
<td>Content analysis of text messages</td>
<td>USA</td>
<td>Children and adolescents 10-17 years with SCD</td>
<td>SCD</td>
<td></td>
</tr>
</tbody>
</table>

1 *HbO refers to haemoglobinopathies which included SCD
Procedure

The studies were read closely several times and key content issues were identified (Step 3). Areas of similarity, overlap and connection were highlighted throughout this process. Following this, key metaphors and themes were recorded which led to the development of a detailed grid (Table 2). This enabled similar issues to be grouped together as ‘key concepts’, marking the first step in determining how the studies were related (Step 4). Second order analyses (Table 3) of the key concepts were then completed. This involved the transformation of key concepts into theoretically informed ‘second-order’ constructs (Schutz, 1962). A third order analysis and the development of a ‘Line of Argument’ then followed, which formed Steps 5 and 6 of the meta-ethnographic approach (Box 1; Noblit & Hare, 1982). The first author was responsible for the preliminary work and fed back provisional results of steps 1 to 6 of the synthesis to the co-authors. An agreement about third order interpretations and a line of argument was reached through discussion of the key concepts and second order analysis.

Results

First Order Analysis: Content issues and key concepts

Table 2 illustrates the completed grid of key content issues from the papers. Concepts are grouped under four abbreviated headings: ‘Informing and being informed’, ‘Preferences’, ‘Functions’ and ‘Consequences’, each of which is defined below:

‘Informing and being informed’: Parents’ or health professionals’ descriptions of their styles and approaches to informing children about SCD. This also includes
children and young people’s experiences of being informed and the dilemmas they face in disclosing their condition to others.

‘Preferences’: Children and young people’s preferences for the way in which they are communicated with.

‘Functions’: People’s underlying objectives when communicating about SCD.

‘Consequences’: Outcomes of people’s communication patterns and styles, as experienced by children and young people.

Concepts and content issues have been contextualised in Table 2 by including brief information about each study sample. Words in quotation marks are original terminology from the empirical papers. This was maintained wherever possible; however, in some cases it was necessary to paraphrase for succinctness.
<table>
<thead>
<tr>
<th>Authors</th>
<th>Participants</th>
<th>Informing</th>
<th>Preferences</th>
<th>Functions</th>
<th>(Negative) Responses of others</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td>Assumptions of child learning from hcps</td>
<td></td>
<td>Normalising</td>
<td></td>
</tr>
<tr>
<td>Gallo et al., (2009)</td>
<td>Parents</td>
<td>Levels of understanding</td>
<td></td>
<td>“Protecting them from being treated differently”</td>
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<td></td>
<td></td>
<td>Forgotten knowledge</td>
<td></td>
<td>Informing others</td>
<td></td>
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<tr>
<td></td>
<td></td>
<td>Understanding not linked to sharing</td>
<td></td>
<td>Understanding and coping</td>
<td></td>
</tr>
<tr>
<td>Gallo et al., (2010)</td>
<td>Healthcare professionals</td>
<td>“Avoid treating condition as secret”</td>
<td></td>
<td>“learning from the experience of living with condition”</td>
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<tr>
<td></td>
<td></td>
<td>“Open and honest”</td>
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<td></td>
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<td>“Starting early on”</td>
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<td></td>
<td></td>
<td>“Matter of a fact”</td>
<td></td>
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</tr>
<tr>
<td>Graff et al., (2012)</td>
<td>Families</td>
<td>Careful consideration of who to share with and what to share Balance in what is communicated by parents</td>
<td>Independence from parents</td>
<td>“Talking to others with SCD”</td>
<td>“(Parents) being over protective”</td>
</tr>
<tr>
<td>Authors</td>
<td>Focus Area</td>
<td>Strategies Implemented</td>
<td>Challenges</td>
<td></td>
<td></td>
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<td>----------------------------------------------------------------------------------------</td>
<td>-----------------------------------------------------------------------------</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Metcalfe et al., (2011)</td>
<td>Parents, Children (8-12 years) &amp; Young People (13-17 years)</td>
<td>Starting early led to better understanding; Assume young children learn from older; Answering questions as they arose; Groups stimulate discussion</td>
<td>Direct information from HCPs needed; Not asking questions to avoid upsetting parents; Not receiving explanations increases stress and strain and could lead to bullying/low self esteem; Fear of upsetting parents-relecatant to ask questions</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Plumridge et al., (2011)</td>
<td>Parents &amp; Children</td>
<td>&quot;Openness&quot;; &quot;Gradual learning&quot;; Small bits over time; Confidence Normalising Less taboo</td>
<td>&quot;Ambivalence about whether or not to declare SCD&quot; Teachers knew but did not understand; SCD not becoming &quot;a central feature framing all life experience&quot;</td>
<td></td>
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<td>Dyson et al., (2010)</td>
<td>Children &amp; young people</td>
<td>&quot;Ambivalence about whether or not to declare SCD&quot;; Teacher knew but did not understand</td>
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<td>Atkin &amp; Ahmad (2001)</td>
<td>Children &amp; young people</td>
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<td>Jacob et al., (2012)</td>
<td>Professionals &amp; young people</td>
<td>Coaching Health promotion Information exchange (hcps)</td>
<td>&quot;Ambivalence about whether or not to declare SCD&quot; Teachers knew but did not understand; SCD not becoming &quot;a central feature framing all life experience&quot;</td>
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Second order interpretation

Second order interpretations were derived from the conceptual grid (Table 1) and are presented under individual concept headings below and summarised in Table 3.

‘Informing and being informed’: The experience of being informed about sickle cell disease and informing others

Consideration of the studies together identified differences in the approaches of parents and healthcare professionals in their communication with children. Some parents (Gallo et al., 2005; 2009) were found to share information selectively, omitting aspects about the condition that they assumed the child would find upsetting. Parents also made assumptions that children were learning about their condition from places aside from the family home, such as from healthcare professionals (Gallo et al., 2005), from older affected siblings (Metcalfe et al., 2011) and from attending support groups (Plumridge et al., 2001) and therefore chose not to communicate with the child themselves. Parents were found to vary in their understanding about SCD (Gallo et al., 2009; Plumridge et al., 2011), with their levels of understanding reportedly unrelated to the amount of information they shared. These communication patterns are suggestive of differences in confidence rather than knowledge, meaning that children may hear incorrect or inaccurate information from parents who are open and confident but do not have a clear grasp of the condition. Parents did however display a lack of confidence about disclosing their child’s SCD to people in the wider community, such as friends or extended family, due to perceptions of stigma and concerns that the child would be labelled.

Healthcare professionals on the other hand were able to deliver consistent, factual information to children and young people and expressed their commitment to
a direct, open and honest communication approach (Gallo, Angst, Knafl, Twomey & Hadley, 2010). Those who followed up children and families in the community (e.g., specialist community nurses), were able to give ongoing practical advice and pragmatic information (Jacob, Pavlish, Duran, Stinson, Lewis, Zeltzer, 2013). Unlike parents, they were unlikely to omit information that they thought children would find upsetting and instead believed it was important to avoid “treating the condition as secret” (Gallo et al., 2010, pp 302).

Comparison of studies that focussed more directly on the experiences of children and young people illustrated the challenges and dilemmas they face in discussing the condition with adults and peers (Atkin & Ahmad, 2001; Dyson et al., 2010, Graff et al., 2012; Metcalfe et al., 2011). A common theme across studies was of the careful consideration of who to talk to and what information to share (Atkin & Ahmad, 2001; Dyson et al., 2010; Graff et al., 2012). Adolescents in particular were found to hold various beliefs about disclosure and communication. Some experienced guilt about the burden they placed on their families leading them to avoid communication or emotional expression (Atkin & Ahmad, 2001; Metcalfe et al., 2011). Some avoided informing teachers and peers, but acknowledged that this came with the disadvantage of other people misunderstanding symptoms or behaviours associated with managing the condition:

I don’t like telling my teachers or my friends. But I feel like, if I don’t, how will they know why I want to go to the restroom or why I need to carry a bottle of water?

(Adolescent with SCD, Graff et al., 2012. pp 327)
Others explained that hiding the condition from others allowed them to maintain the impression of normality, for example:

I try to ignore it most of the time. That’s why I don’t tell people. It’s like I’m just trying to get away from it. I’m just trying to pretend that I’m normal.


Preferences about the style and approach to communication about SCD

As discussed above, parents use various strategies to protect their child from hearing potentially distressing information (Gallo et al., 2009) or to avoid talking about SCD altogether (Metcalfe et al, 2011). The analysis has however revealed that children, especially those between the ages of eight and twelve, express positive feelings about talking and are keen to hear about all aspects of their condition. Open and honest communication with people who show understanding and empathy also appeared to be important for them. They described this happening in their discussions with other affected children (Jacob et al., 2013; Plumridge et al., 2011) and with healthcare professionals (Atkin & Ahmad, 2001; Metcalfe et al., 2011; Jacob et al 2013) but not parents. A study of wireless “text” communication using mobile phones between young people and healthcare professionals reported that many participants requested to be connected with other young people in the study for emotional and psychosocial support (Jacob et al., 2013). Children and young people valued gradual learning approaches (Metcalfe et al., 2011) and believed that attendance at SCD support groups, facilitated by healthcare professionals, would
enable this (Plumridge et al., 2011). Discussions with healthcare professionals were highly valued by many children and young people who felt that they had a more complete understanding compared to parents, especially concerning genetic risk information (Metcalf et al., 2011). They also felt they were likely to share information openly and honestly (Gallo et al., 2009). Adolescents also reported having discussed friendships and romantic relationships with healthcare professionals, and described how this level of informal social support acted to strengthen therapeutic relationships (Atkin & Ahmad, 2001; Jacob et al., 2013).

As children grow older and reach adolescence, the need for independence in managing their condition appeared to grow. Reminders from parents about taking medication and keeping hydrated became increasingly viewed as “overprotective” and deemed unnecessary in their efforts to adapt and manage SCD independently (Graff et al., 2012). Whilst younger children with SCD appear more accepting of parental protection and support, adolescents were found to begin asserting their independence by confronting their condition and challenging the limitations imposed on them (Atkin & Ahmad, 2001). Some young people wished to live a normal life that was not framed by SCD (Atkin & Ahmad, 2001; Dyson et al., 2010). Others were more willing to accept their condition and wanted to begin taking more responsibility for managing their symptoms, something that they struggled to communicate to parents. In a focus group discussion, one adolescent asked for advice about an ‘overprotective’ parent and was given the following advice by other respondents:

Adolescent 1: …let her let you live because you learn how to be your own person
Adolescent 2: And you learn to take care of yourself.

(Graff et al., 2012, pp 330)
Talking to others about SCD had a number of functions for children and young people including educating peers about their condition and helping people understand illness related behaviours and school absences (Dyson et al., 2010; Jacob et al., 2013). Some described the benefit of offloading to parents to cope with negative feelings and distress (Atkin & Ahmad, 2001) and informing others about their symptoms to receive appropriate care (Dyson et al., 2010). Parents believed that their communication functioned to normalise the condition whilst promoting positive adaptation (Gallo et al, 2005); however, the analysis suggested that despite their intentions, their communication is not always conducive to promoting adaption. Selective sharing of information about SCD within and outside of the family (Gallo et al., 2009) is one example. Another was of downplaying the condition by comparing it to more ‘serious’ diseases such as cancer or more severe forms of SCD, such as “…those that required transfusions or those where strokes had occurred” (Gallo et al., 2005., p 272).

Healthcare professionals described a similar goal of promoting adaptation in wanting to help children “…understand and cope with symptoms and better prepare them to learn from the experience of living the condition” (Gallo et al., 2010, pp 302). Their communication was aimed at promoting independence and self-management through collaboration and coaching around the physical and emotional aspects of the condition (Jacob et al., 2013). In contrast to parents, they described a consistently open and honest approach, one that promoted positive adaptation and independence for young people affected by SCD.
Consequences of communication patterns in SCD

As previously discussed, young people, especially adolescents, may experience their parents’ guidance as unnecessarily overprotective in their efforts to gain independence (Graff et al., 2012). Younger children were reluctant to discuss SCD with parents due to feelings of guilt about the distress their illness had caused for family members as one young female (aged 13) with SCD explains:

She gets so upset when I’m having a pain and it’s not fair on her, like I don’t want to make her worry. Because if she’s upset I get upset for upsetting her.


Atkin and Ahmad (2001) write that, “It was important to cope, for their [family members’] sake” [pp 620]. In such cases, where there was avoidance of parental upset or when parents had not acknowledged the genetic condition openly, children were reluctant to ask the questions that may have resulted in better understanding and coping with their condition (Metcalfe et al., 2011).

Feelings of isolation and experiences of bullying were common in the reports of young people (Atkin & Ahmad, 2001; Dyson et al., 2010; Metcalfe et al., 2011) with closed family communication suggested as one contributing factor (Metcalfe et al., 2011); however, the precise mechanism of this has not yet been explored. One possibility is that without accurate and positive family communication, young people become fearful of a condition that they do not fully understand, leading to low self-esteem and social withdrawal. Conversely, those who felt they had a good understanding were more likely to help peers and authority figures, such as school
teachers gain a shared sense of understanding and acceptance of the condition (Dyson et al., 2010; Jacob et al., 2013).

Teasing from others was frequently reported by young people with SCD (Dyson et al., 2010; Atkin & Ahmad., 2001). Young people reported bullying and teasing that was experienced as “negative in an ignorant way” by peers and teachers [Dyson et al., 2010. pp 2041]. One young person made the analogy of SCD being ‘the monster in the closet’ in that those without an understanding of the condition may not know “…what the facts are and if someone doesn’t know, [they] might get scared of it” (pp 2041). In some cases, others were aware of SCD but did not understand the health related needs of an affected child (Dyson et al., 2010). This often resulted in them being ignored, punished or mocked by other pupils and teachers and had implications for children’s self-esteem and academic progress (Atkin & Ahmad, 2001).

*Developing a line of argument: Third Order Analysis*

Interpretations derived from the meta-synthesis reveal a miss-match in the communication styles of parents and healthcare professionals caring for children with SCD. This may result in children hearing mixed messages about the acceptability of talking about their condition and a consequent ambivalence about disclosure. Their communication styles are likely to become maladaptive without appropriate modelling from parents. For those who chose not to disclose in school environments, subtle signs such as tiredness, poor concentration, needing to go to the toilet often, do not go unnoticed by peers or teachers who were found to respond in “ignorant” and discriminatory ways. These responses, or a fear of them, may further inhibit communication as children act to protect themselves from unwanted attention.
or bullying. Conversely, in times where communication about SCD is open, clear and consistent, children were more likely to discuss their condition in school, with teachers and peers. This open communication offers to empower children, and help them promote the acceptance, understanding and empathy they yearn for in others, thereby offering to enhance wellbeing and quality of life.
Table 3. Second and third order analyses

<table>
<thead>
<tr>
<th>Key Concepts</th>
<th>Second order analyses</th>
<th>Third order analyses</th>
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<tr>
<td>Informing others</td>
<td>Parents have varied understandings, can be selective in what they share and make assumptions that children will learn from elsewhere. Healthcare professionals communicate pragmatically, sharing information with children openly and honestly. Children and young people experience dilemmas in what they should disclose and to whom.</td>
<td>To tell or not to tell…. Children receive mixed messages about the acceptability of communication about SCD leading to ambivalence about disclosing SCD to others. Facilitating Adaptation Open communication is critical for developing functional adaptation. Having confidence in communicating openly about SCD promotes the honesty and openness that young people desire. Children need clear consistent messages within open communication from early on for this to happen.</td>
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<tr>
<td>Preferences</td>
<td>Children want open communication with people who can understand and empathise with them. They value talking with HCPs and other young people with SCD.</td>
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<tr>
<td>Functions</td>
<td>Effective communication offers to promote functional adaptation i.e. learning to self-manage whilst maintaining a sense of normality.</td>
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<tr>
<td>Consequences</td>
<td>Young people’s avoidance of open communication about SCD can to lead to isolation and discrimination.</td>
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Discussion

By taking the perspective of the child with SCD, this meta-synthesis has provided insight into how children are communicated with and how they may experience this communication. Children were found to communicate with their families, peer groups, teachers and care teams. However, the content and style of communication from each of these groups was noticeably different. The incongruous way in which this communication co-occurs is considered as a contributing factor in the ambivalence children and young people described about disclosing their condition to others. Notably, ambivalence increased with age, which may reflect a stage in the life course where young people begin negotiating their identity and place in society (Crocket & Silbereisen, 2000). However, confounding messages about their condition are likely to impact negatively on this process. Indeed, young people with chronic conditions have been found to experience difficulties negotiating adolescence, as the protection of their long term health conflicts with their current priority of gaining social acceptance as well as vocational goals (Yeo & Sawyer, 2005).

Parents’ use of avoidant communication strategies, such as communicating selectively or limiting communication about SCD, was a key finding of the synthesis. Some of their reasoning for this included the assumption that children would learn about their condition from elsewhere. This strategy risks children misunderstanding their condition and having a poor sense of illness-related identity. Communication could also be limited due to parents’ concerns about their child being ‘different’. Given these anxieties, it is no surprise that some adolescents described an intense desire to be ‘normal’. Overall though, the responses of young people differed, reflecting previous findings of markedly varied coping responses of
young people with SCD, which are likely to be influenced by parental approaches and family environment (Kliwer & Lewis, 1995).

It is important to consider the sociocultural influences which may affect children’s and young people’s coping. SCD is an incurable illness affecting minority ethnic groups and one with associated stigma based on uneducated notions about the nature and course of the disease (Burnes, Antle, Williams & Cook, 2008). Whilst the negative experience of stigma associated with SCD is likely to be shared by other family members, parents and extended family may themselves have disablist attitudes (Atkin & Ahmad, 2000), thereby maintaining a ‘culture of silence’ within BME communities (Burnes et al., 2008). The finding that support groups helped parents overcome a sense of “taboo” in discussing SCD (Plumridge et al., 2011) is undoubtedly a positive function of the groups. Of equal importance though is parents’ indirect admission here that talking about SCD with their child feels uncomfortable. Thus, facilitating conversations away from the home initially may be the first step in helping children and parents talk more openly about SCD. Recently, a study of multi-family discussion groups (MFDG) has provided evidence for the feasibility of MFDG interventions to improve condition related communication in families affected by a genetic condition or genetic risk (Eisler, Ellison & Flinter, 2015). Research into this style of intervention in SCD would be of benefit to children, families and associated professionals. Interventions such as this may offer a joint role for sickle cell nurse specialists and psychologists to reduce feelings of family discomfort and facilitate more open communication that promotes acceptance and functional adaptation to SCD.

Conclusions about children’s lived experiences of parental communication could not be drawn from many of the studies. However, it is possible that approaches
that involved minimising the condition in an attempt to normalise it, could leave a child feeling invalidated and unable to express their emotions. Conversely these approaches may be experienced positively by the child, as their parents attempt to reframe the illness experience, which has been showed to promote active coping in children (Kliewer & Lewis, 1995). It was however healthcare professionals and others with SCD, not parents, who were described as the most supportive suggesting that parental approaches may not have been experienced in the way they intended.

**Limitations**

The meta-synthesis is not without limitations. Several papers were included that presented findings on a single sample, which could be criticised for providing a limited pool of participants to base findings upon. With this in mind, inclusion of these studies was carefully considered from the outset. The decision to include all papers from the same samples was based upon findings of similar and co-occurring themes when the papers were compared with findings from other samples. In addition, our intention to maximise the inclusion of SCD related studies in a relatively sparse field of literature.

The inclusion of papers that reported findings on a large range of genetic conditions may also be considered a limitation in that SCD specific issues may not be adequately explored or captured. This was considered during the screening process, where findings sections were scrutinised to ensure that data related to SCD could be differentiated from those concerning other conditions.

Finally, conclusions about a child’s lived experience of communication about SCD cannot be made from this study and only their experiences of co-occurring but incongruent communication can be inferred. Future research would benefit from
investigating children’s perceptions of these messages and the impact it has on their confidence to disclose to peers and healthcare professionals in times of crisis. Indeed, Derlega, Janda, Miranda, Chen, Goodman and Smith (2014) suggest that previous experience of open communication with others affected by SCD correlates with a willingness to approach healthcare professionals to obtain appropriate care during crises. Predictive models, using a prospective design are therefore indicated to further investigate this relationship.

Conclusions and recommendations for practice
This is the first meta-synthesis of studies related to communication in SCD and has allowed us to consider issues specific to SCD that may not be possible when studying SCD within a broader range of genetic conditions. Communication that is tailored to the child’s developmental stage, is consistent across family and professional groups and which coaches the child to manage independently are known factors in helping children adapt to their condition (Metcalfe et al., 2008). This review highlights these issues and confirms existing findings that parents may need support to ensure that they are able to communicate effectively with their children about SCD. It also adds information about the range of messages children and young people hear from different personal and professional forums, and the impact this may have on their identity and adjustment. It is recommended that children are encouraged to talk openly about SCD with parents, healthcare professionals and both affected and unaffected peers. Parents should be modelling acceptance of SCD in the family, whilst providing children with knowledge and skills to communicate openly; however, they may require professional support with this. Healthcare professionals working within specialist SCD services are well placed to provide this support given
the nature of the relationship they develop with families. Provision of quality training and continuing professional development (CPD) for healthcare professionals in facilitating open family communication about SCD should therefore be on the agenda of commissioners and SCD service managers.
References


Atkin, K., & Ahmad, W. I. (2001). Living a ‘normal’ life: young people coping with thalassaemia major or sickle cell disorder. Social Science & Medicine, 53(5), 615-626


NHS Sickle Cell and Thalassaemia Screening Programme (2010) url:


Communication with children about sickle cell disease: A qualitative study of parent experience

Paper 2 has been prepared for submission to Qualitative Health in accordance with the guidelines for contributors Research (Appendix 3).

Word Count: 7981 (Excluding references)

Abstract Word Count: 148
Abstract
This study aimed to explore parents’ experiences of communication with children affected by sickle cell disease. Semi-structured interviews were conducted with twelve parents whose child had a diagnosis of sickle cell disease. Interviews were audio recorded, transcribed verbatim and analysed using inductive thematic analysis. Themes related to four areas of communication: pre-conditions facilitating communication, proactive coaching styles of communication, avoidant communication, and the impact of communication on the child’s knowledge, role and relationship with the parent. ‘Coaching’ styles of communication reportedly helped children negotiate the challenges associated with managing sickle cell disease. Descriptions of avoidant communication were also common, often driven by fears of their child’s ability to cope with the truth. The findings suggest a need for healthcare professionals to support parents in openly communicating and overcoming barriers to communication about difficult topics. This would facilitate consistency in communication between parents and healthcare professionals, thereby enhancing child adjustment.

Key Words: sickle cell disease, SCD, communication, parent-child communication, qualitative analysis, thematic analysis
**Introduction**

Previous research in paediatric chronic illness has identified that children who are included in discussions about their condition cope more effectively than those who are not (Cohen, 1999). Other research has identified the negative impact of poor communication on a person’s illness management and adjustment (Rosland, Heisler & Piette, 2012). Theories of illness perceptions and behaviour suggest that illness related knowledge and beliefs are influential in the adoption of preventive health measures as adults (Leventhal, Safer & Panagis, 1983). A child’s socialisation to their condition is a key period in the development of health and illness beliefs (Kliwer & Lewis, 1995), within which parental communication and behaviour is influential (Koopman, Baars, Chaplin & Zwinderman, 2004). Parental patterns of communication lead to the gradual development of positive or negative health beliefs that influence autonomy and self-management of illness (Rosland et al., 2012).

Genetically inherited chronic illnesses, such as Sickle Cell Disease (SCD) present families with additional challenges, when feelings of responsibility, guilt and cultural stigma have the potential to impact on the way in which children are communicated with (Thomas & Taylor, 2002). Maladaptive styles of coping are apparent in adults with SCD (Maxwell, Streetly, & Bevan, 1999) and theories of illness beliefs suggest that they are developed early on through social learning and parent-child communication (DiMatteo, Haskard & Williams, 2007).

*Sickle Cell Disease*

Sickle Cell Disease (SCD) is a lifelong hereditary blood disorder, most commonly affecting people of Black and Minority Ethic (BME) origin (De, 2005; Graff, Hankins & Graves et al., 2012). It is one of a number of blood disorders, including
Thalassaemia Major, known collectively as ‘Haemoglobinopathies’. In SCD, abnormally shaped red blood cells cause small vessel blockages that account for a number of physical symptoms (often pain related) and long term organ damage (De, 2005). Affected children have chronic anaemia, impaired growth, episodic painful crises and many suffer life threatening complications, including haemorrhagic stroke (Sheth, Licursi & Bhatia, 2013). Various forms of sickle cell disorders exist, the most common being homozygous SCD (HbSS), which is associated with more frequent and severe symptoms. Other types include HbSC disease and Sickle Beta Thalassaemia (HbSβThal) which are associated with more moderate symptoms but require similar levels of self-management and monitoring by specialist teams.

Significant progress has been made over the past century in managing SCD with medication (Hydroxycarbamide), prophylactic antibiotic treatment and, in limited cases, stem cell transplantation. Despite these improvements, life expectancy remains significantly reduced compared to the normal population, with UK estimates of between 53 and 58.5 years for men and women, respectively (NHS Sickle Cell and Thalassaemia Screening Programme, 2010).

**Psychological adjustment in SCD**

Historically, the unpredictability of physical symptoms and a reduced sense of control over the progression of SCD have been found to impact on the psychological

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2 Sickle cell anaemia (HbSS) occurs when the haemoglobin variant (HbS) gene is inherited from both parents. When the HbS gene is inherited from one parent and another haemoglobin variant gene from the other parent, this leads to different types of the disease such as sickle cell haemoglobin C disease (HbSC) or sickle beta-thalassaemia (HbSβThal). Sickle cell trait (HbAS) is the the ‘carrier’ state, and is inherited through the HbS gene from one parent and a normal haemoglobin (HbA) gene from the other (Anie, Steptoe & Bevan 2002). A child of carrier parents has a one in four chance of inheriting the condition.
wellbeing of affected adults (Lewis & Kliewer, 1996). Psychological difficulties, such as anxiety and depression, have been identified in a proportion of children and adults with SCD (Benton, Ifeagwu & Smith-Whitley, 2007; Levenson, McClish & Dahman et al., 2008). Those who experience psychological distress have also been found to have high levels of negative thinking and passive coping styles, which have been linked with more severe symptoms, unhelpful pain management strategies and increased incidence of emergency care (Anie, 2005; Gil, Abrams, Phillips & Keefe, 1989; Gil, Abrams, Phillips & Williams, 1992).

Health related quality of life (HRQL) in SCD has been widely studied. Adults and children with SCD have been found to have a significantly reduced HRQL compared to healthy individuals and comparable or worse HRQL than those with other chronic diseases (Panepinto & Bonner, 2012). However, a recent UK study that used an alternative quality of life (QoL) measure to compare children with SCD with healthy controls found that ratings did not differ significantly (Constantinou, Payne & Inusa, 2015). This measure asked children to rate aspects of their current life against their imagined ideal. The authors suggest this finding may reflect children’s expectations being in line with the limitations associated with their condition. Children had therefore developed realistic expectations and aspired to achieve within their limits. The sample size in this study was small, which not only limits generalisability but may also reflect a sampling bias in that those who took part came from families who were open to communicating about their wellbeing. It is therefore likely that open communication about SCD is an important factor in facilitating child acceptance and adjustment; however, this has been insufficiently studied in SCD research.
Parent-Child Communication in SCD

A number of factors have been suggested that may influence the way in which a child with SCD is communicated with. Anxiety, overprotection, guilt and denial have been found in parents of children with genetic conditions (Whitten & Fischhoff, 1974; Graham, Reed, Levit, Fine & Medalie, 1982; Ulph, Cullinan, Qureshi & Kai, 2011) and in SCD specifically, have been linked with avoidant communication styles (Gallo, Angst, Knafl, Hadley & Smith, 2005; Middleton, Ulph & Calam, In Preparation; Thomas & Taylor, 2002). Family environments and parental coping styles are known factors in the development of illness beliefs and coping strategies in SCD (Kliwerer & Lewis, 1995). Thus parental knowledge, beliefs and perceptions of a child’s illness are likely to influence communication patterns and a child’s socialisation to their condition. In an investigation of parental understanding about child prognosis in SCD, Roth, Krystal, Manwani, Driscoll and Ricafort (2012) identified a considerable gap in parental awareness of the reality of their child’s prognosis and the likelihood of future limitations. Studies of chronic illness more generally have described parental uncertainty of a child’s prognosis as a barrier to accurate and honest communication (Rolland, 1994). Finally, illness related stigma in SCD is has been well documented (Burnes, Antle, Williams & Cook, 2008; Bediako & Moffitt, 2011; Jenerette, Funk, & Murdaugh, 2005; Thomas & Taylor, 2002) and may pose an additional barrier to open parent-child discussions.

Given these challenges, parents are likely to benefit from support to overcome barriers to communication. Healthcare professionals are well placed to provide this type of support, which has been defined as “…practical assistance, positive role modelling, and emotional, informational, and affirmational support” (Letourneau, Drummond, Fleming, Kysela, McDonald, Stewart, 2001, pp 163). Support has also
been conceptualised by parents as three dimensional - including counselling, instrumental or practical help, and information provision (Kirk & Glendinning, 2002). Through an in-depth exploration of parent experience, this study sought to provide recommendations for ways in which parents can be supported to improve communication with their child and manage the difficult feelings that may occur in the process.

Aims

SCD is often studied alongside, that is, as part of a larger sample of genetic conditions. As previously argued (Middleton, Ulph & Calum, In preparation), SCD presents specific issues relating to condition management and culture, thereby warranting individual study. Very little research exists that explores the experience of parents of younger children with SCD, who may be facing unique challenges in socialising their child to the condition and tailoring information to their stage of cognitive development (Koopman et al., 2004). Existing studies have also tended to be completed in non-UK countries (e.g., USA, Nigeria) and thus may not represent the experience of parents living in the UK. Finally, existing UK studies of parents and families affected by SCD have tended to focus on communication about genetic risk rather than about ongoing symptomatic and management issues. This study aimed to address these gaps by asking parents about their experiences of communicating about SCD and their needs for support in this process.

The following research questions guided the investigation:
1. What are the experiences of parents in communicating with their child about SCD?

2. What support do parents need for communicating effectively with their child about SCD?

Research questions were broad with the aim of gathering rich information about parental experiences. The ontological approach was grounded within the theoretical position of contextualism. The contextualist approach sits between the poles of realism and constructionism and considers knowledge as local, provisional and situation dependent (Jaeger & Rosnow, 1988). Understandings of experience are thus considered to be influenced by the broader social context, but still considered valid representations of individual experience.

Methods

Ethical Approval

Ethical approval was granted for this study by Solihull Health Research Authority, NHS Research Ethics Committee (REC reference: 14/WM/1093) (Appendix 4). Approvals were also gained from the Research and Development department of the NHS Foundation Trust participants were recruited from (Appendix 5). Data were anonymised at the point of transcription. Recruitment and data collection were completed by the lead author, who was the principle researcher of this study.

Participants

Inclusion criteria were parents of children aged 7 to 14, with a diagnosis of any sickle cell disorder. In total, 12 parents were recruited from a paediatric haemoglobinopathy clinic at a children’s hospital in the North of England. Parents of
children with other haemoglobinopathies (e.g. Thalassaemia Major) were excluded due to the incomparable symptomatology and condition management. Parents who met inclusion criteria were identified by a field researcher who was a Clinical Psychologist working within the Sickle Cell and Thalassaemia Service. With the aim of gaining a rich and varied insight, sampling was purposive to ensure that the study included a diverse group of participants with ranges in child age, type of SCD and parent characteristics, such as gender, employment status, marital status and education. Participant demographic information is shown in Table 1.

Table 1. Participant Characteristics

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<td>Child age</td>
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</tr>
<tr>
<td>High school</td>
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<tr>
<td>Further education</td>
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<td>Graduate</td>
<td>7</td>
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<tr>
<td>Employment:</td>
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<td>Full time</td>
<td>4</td>
</tr>
<tr>
<td>Part time</td>
<td>3</td>
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<tr>
<td>Student</td>
<td>1</td>
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<tr>
<td>Not working</td>
<td>5</td>
</tr>
<tr>
<td>Marital status:</td>
<td></td>
</tr>
<tr>
<td>Married</td>
<td>9</td>
</tr>
<tr>
<td>Separated/Single</td>
<td>3</td>
</tr>
</tbody>
</table>
**Procedure**

Eligible parents were approached in the clinic waiting room. Parents were provided with a verbal overview of the study and offered the opportunity to discuss it in greater detail with the principal researcher. Parents who agreed were introduced to the researcher who handed them a Participant Information Sheet (PIS) (Appendix 6) and a ‘Consent to Contact’ form (Appendix 7). Those who signed the form were contacted by the researcher, who outlined the aims of the study and provided the opportunity for parents to ask questions. Face-to-face interviews were conducted with those who gave informed consent (Appendix 8) to take part and parents were given the choice of completing the interview at home or at the Manchester Sickle Cell and Thalassaemia service. Interviews lasted between 40 and 90 minutes and the majority (9/12) took place in participant homes. Out of the remaining three, two were conducted at the local Sickle Cell and Thalassaemia Centre and one was conducted on the telephone. Interviews were conducted individually by the principal researcher who obtained informed consent to take part and ensured participants were aware of their right to stop the interview at any time and withdraw from the study. Interviews were audio recorded and transcribed verbatim throughout the interviewing phase.

**Measures**

A semi-structured interview schedule was developed by the principal researcher, with support of the research team. The team included academic and clinical professionals who had expertise in qualitative methods and genetic condition research (FU), child and family research (RC) and psychological issues in SCD (SP). The team provided feedback resulting in the development of the final version
(Appendix 9). Questions were open ended but supplemented with prompts to promote broad understandings of parental experience. The schedule was informed by literature on family communication about genetic conditions and was adapted to suit parent-child communication in SCD. To enhance rigour, a pilot interview was conducted with a SCD parent support group member who was a mother of a 7 year old daughter with SCD (HbSS). In the pilot interview, the researcher interviewed the pilot interviewee using the drafted interview schedule. This process enabled the researcher to assess the comprehensibility and flow of the schedule and involved asking the pilot interviewee for feedback on the questions in the schedule. The pilot participant suggested additional prompts to follow questions about the first conversation to help people remember what had been said. The addition of a questions about ‘what is not talked about’ was also suggested due to the pilot interviewee’s shared belief that parents were likely to have either deliberately or unknowingly avoided some topics. Questions about ‘not talking’ were consequently added to the schedule, for example “What haven’t you talked about?” and “What are your reasons for not talking about X”. Having become familiarised with the adapted schedule, interviews were completed using a condensed version (Appendix 10). This enabled greater flow, flexibility and rapport during the interviews and in turn promoted more complex and meaningful interpretations of the data. Demographic details of participants were collected through a participant questionnaire (Appendix 11), which was administered before the start of each interview.
Data Analysis

An inductive approach to analysis was employed to ensure understandings of communication remained centred around the shared meanings and experiences of parents (Braun & Clark, 2014). Thematic analysis was chosen based on the flexibility it allows in exploring participant experience, whilst facilitating rich and complex interpretations of the data (Braun & Clarke, 2006). The analysis adhered to the seven stages of thematic analysis recommended by Braun and Clark (2006) which are listed in Box 1 and described in full below.

A reflective journal was kept throughout data collection and analysis to promote a reflexive stance, which has been claimed to enhance quality and credibility (Armstrong, Gosling, Weinman & Marteau, 1997; Ortlipp, 2008). Personal reflections and feelings related to arising issues were recorded to promote transparency of the influence of personal beliefs and values on the collection or interpretation of data (Watt, 2007). The researcher’s position as a white female and status as a parent were considered to have the potential to influence the data owing to both shared and different cultural and parental experience. The researcher’s sense of shared experience is likely to have promoted an empathic stance and thereby facilitated more valid interpretations of the data. The differences in ethnic and cultural backgrounds, however, may have negatively affected interpretative validity (Ortlipp, 2008).
Interview recordings were transcribed verbatim throughout the interviewing phase (Stage 1). The researcher became familiar with the data by thoroughly reading each transcript. Data collection was paused half way through the interviewing phase, at which time the researcher reflected on the existing demographic of the sample and common discussion points, that is items of ‘potential interest’ (Stage 2). This was discussed among the research team and led to a more purposive recruitment strategy for the remainder of the study to ensure a varied parent and child demographic was included. It also led to more directive questioning about parent coping and perceptions of their child’s understanding.

In line with stage 3, data were coded completely, that is, transcripts were read systematically and data relevant to the research questions were assigned a descriptive code.

Coding was completed electronically using QSR Nvivo (Version 10). Codes were reviewed by the principal researcher half way through the coding process and again on completion, where each code was considered according to its relevance to the
research questions. This led to irrelevant codes being separated and codes with similar content being merged. A pattern based analysis was then started (Braun & Clark, 2013) in which groups of related codes which were clustered together and collapsed to form candidate themes (Stage 4). At this stage, the research team met to discuss the emerging thematic framework, at which point themes were reviewed against the coded data and the whole data set in relation to the research questions. Having agreed on four main themes a thematic map was developed (Stage 5) showing each of the themes and the final condensed codes within each theme (Figure 1). Themes were then named and defined leading to the final stage of writing up the analysis.

Findings

Four key themes were identified through the process of inductive thematic analysis which were: Pre communication conditions (Theme 1), Proactively coaching via communication (Theme 2), Avoidance of communication: “Don’t dwell on the negative” (Theme 3) and Outcomes of communication (Theme 4). Themes are described below and are supplemented with anonymised data extracts. Figure 1 illustrates the themes diagrammatically.
Figure 1. Theme diagram

- **Theme 1. Pre-communication conditions**
  - Child curiosity
  - Parental attunement

- **Theme 2. Proactively coaching via communication**
  - Active coaching
  - Indirect coaching

- **Communication experiences**

- **Theme 3. Avoidance of communication: “Don’t dwell on the negative”**
  - The impact of stigma
  - Protecting wellbeing

- **Theme 4. Outcomes of communication**
  - Knowledge
  - Role
  - Relationship
Theme 1: Pre-communication conditions

Prior to any discussion between a parent and child about SCD, a number of pre-existing conditions were identified that were necessary prerequisites of communication. These related to the parent in their ‘attunement’ to their child’s needs and to the child, who often triggered communication in response to their own curiosity about aspects of the condition.

Parental attunement

Maintaining an awareness of child wellbeing, understanding and informational needs was a key facilitator of open communication about SCD. Parents described ongoing monitoring of children’s physical health and mood. As one parent explained:

‘We are putting our eyes open every single day’.

(P12, Father of daughter, 13, HbSS).

This high level of attentiveness was found to prompt exploratory communication aimed at identifying the source of a child’s distress:

‘Yeah sometimes when she is quiet, I know something is going wrong. I will bring her closer. “What is going on?” “Do you have pain?” “Do you have pain at school?” “What happened at school?” “Did anybody hurt you?”…’

(P3, Mother of daughter, 7, HbSS)
Maintaining this level of awareness, whilst described as a challenge, was something many parents appeared to be doing intuitively. One difficulty, however, was of judging when and what to communicate:

’I think the part that I found really difficult was working out…what age do you tell them? What is too much information?’

(P7, Mother of daughter, 10, HbSS)

Careful consideration of this dilemma was suggestive of their attunement to their child’s needs. Furthermore, despite some parents concerns, many claimed to be tailoring information to their child’s developmental stage.

_Child curiosity triggers communication_

The occurrence of symptoms, medical procedures or social events had the potential to trigger communication about SCD most often originating from a child’s curiosity and questioning. Children were also found to initiate early conversations by posing questions about symptoms:

‘…[Daughter asks] “Why does it hurt? Why is my hand swollen?”… I’d say it’s because of your blood. At that time all she understood was ‘my blood’ But then as she got older, coming for appointments then I thought she’s in a better position to grasp it.’

(P1, Mother of daughter,10, HbSS)
Parents commonly described giving children vague responses to younger children (e.g. under 7 years old); however, as the parent above suggests information was expanded on as the child aged to meet their level of understanding and readiness.

Curiosity about care and management practices imposed by parents also triggered communication for example asking: “Why do I have to drink water?…” “Why do I have to keep warm?” (P1, Mother of daughter, 10, HbSS). Others spoke of their child’s exploration about ‘Why’ they had the condition, something that many parents described as a challenge to answer, not for lack of knowledge but related to their sense of responsibility:

‘So she was asking me why is she [name of genotype]. So it was then that I told her, that I didn’t know that your father was a carrier. I myself I didn’t know, until I gave birth to you’.

(P3, Mother of daughter, 7, HbSS)

In some cases, it was clear how a combination of parental attunement and child curiosity led to open and effective communication. For example, staying closely attuned meant parents were more able to notice opportunities to talk as P10 describes:

‘It’s just finding that balance…if he starts showing an interest it’s easier for us to talk to him about it, because he asks me question and I use that opportunity to really talk’.

(P10, Mother of son, 13, HbSS)
Theme 2: Coaching through communication

Many parents described positive communication styles in which they had communicated openly and proactively with their child about managing SCD. They described having discussed a range of topics, shown in Table 2, related to medical and social issues. Parents spoke about the first conversation they had with their child about SCD and, when prompted, about topics that they had avoided or excluded from conversations.

Table 2. Communication topics

<table>
<thead>
<tr>
<th>Medical</th>
<th>Social</th>
</tr>
</thead>
<tbody>
<tr>
<td>Physiology (Blood)+</td>
<td>Informing child of difference+</td>
</tr>
<tr>
<td>Genetic inheritance+</td>
<td>Informing child of limits+</td>
</tr>
<tr>
<td>Pain+</td>
<td>Decision making</td>
</tr>
<tr>
<td>Bone marrow transplant*</td>
<td>Regret</td>
</tr>
<tr>
<td>Appointments &amp; procedures</td>
<td>Normalising SCD</td>
</tr>
<tr>
<td>Potential complications*</td>
<td>Offering hope</td>
</tr>
<tr>
<td>Death*</td>
<td>Inheritance (e.g. choosing a marital partner)*</td>
</tr>
</tbody>
</table>

+ Common first conversation topics
*Topics found to be commonly avoided

Proactive coaching

Underlying many of the topics shown in Table 2, however, was parents’ intent to ‘coach’ their child about SCD so as to promote understanding and self-management. Parents were found to be focussing on equipping their child with the knowledge and skills to become competent and confident in managing their condition. Many described their attempts to positively reinforce aspects of a child’s condition.
management. The parent below describes a typical conversation around management regimes:

‘I will encourage her to eat ... [as if to the child] “If you don’t eat you will be getting weaker and weaker” ... “And then you need to take plenty of fluid”. [Child says] “Oh no why? I don’t want to because if I take too much fluid I want to go to toilet every second”. I say “Yes, that is true, I know. But you have to keep going, giving you strength, and keeping you happy. Then when you are playing with your friends you have more energy to carry on’.

(P7, Mother of daughter, 10, HbSS)

This motivational approach to coaching aimed to promote self-management by focussing on the longer term outcomes both generally, i.e. giving strength, and specifically, in tailoring the proposed outcomes to their child’s values.

Regarding medication, parents reacted to their child’s reluctance with encouragement using various approaches. One such approach involved reinforcing the long term effects:

‘But each time she doesn’t want to take, I say “Hey, this medicine you have to take it. You know you are sickle cell. If you miss it now it’s not going to work … take it, you will see you will not have pain as much.” She will say “okay”…’

(P3, Mother of daughter, 7, SCD, HbSS)
Reluctance or nonadherence was also reacted to with communication about the potential for serious consequences as in this example:

‘If you don’t take [medication], it can cause a serious thing. [Daughter says] “What serious thing is that?” So I tell her it can result in death, where you don’t [sic] be in the midst of the family again.’

(P5, Mother of daughter, 12, HbSS)

Another approach was to offer praise in order to reinforce the child’s self-management efforts and encourage a sense of ownership and control in managing the condition:

‘The consultant said it was brilliant, there was no iron or anything, so then I said, “See, because we’ve doing really good with the [name of drug]”, [I] remind him… that’s why we have to take this so you’ll always get a good result.’

(P6, Mother of son, 9, HbSS)

This could be followed by a problem solving discussion to:

‘…try to see what could have triggered it and how we could do it differently next time’.

(P6, Mother of daughter, 7, SCD, HbSS).
Others, however, approached this in a more disciplinary manner:

‘I said “What happened?” She said was doing P.E. She didn’t remember to put on her tights. Then after that she was having pain. I said “Okay. But how many times have I told you that when you are going out, put on your coat, put on your tights so that you don’t catch cold”. She said “Mummy…” [pause]. I thought good - don’t make that mistake again.’

(P3, Mother of daughter, 7, HbSS)

Discussions from hospital appointments were often continued at home and tended to be centred around medication. They were also opportunities to offer clarity and reassurance about potentially upsetting information about prognosis. For example:

‘If I hear anything like that I’ll say to her afterwards “That doesn’t apply”. ‘Don’t worry about that because it doesn’t apply to you yet. It may do in the future but don’t take it on board yet.’

(P8, Father of daughter, 11, HbSC).

Coaching about genetic inheritance was common and often involved informing children about different types of gene patterns associated with SCD and how transmission can be prevented when they have children of their own. This was particularly true for parents of children with the HbSS genotype, for example:
‘And then I told him if you want to get married, I told him you have to do a blood test before you get married to the person so you see … if [they are] compatible so that you don’t have SS in future.’

(P4, Mother of son, 10, HbSbThal)

Parents of children with HbSC Disease or Sickle Beta Thalassaemia rarely reported having had these discussions, perhaps reflective of the less severe symptoms associated with these conditions. Those with HbSS however were made fully aware, from an early age, of how SCD is inherited and the way in which their lives may be influenced by the condition. Children’s abilities were often compared to unaffected others, highlighting the difference between them and healthy children. This was often communicated to them through discussions about exercise or play, for example:

‘I know it’s exciting but it’s not good for you”. She goes on to discuss how importance of: “making her come to terms again with the fact that she is just a bit different from some of her friends.’

(P3, Mother of daughter, 7, HbSS)

*Indirect coaching*

Non-verbal and indirect forms of communication appeared to be aimed at conveying warmth, understanding and positive messages about SCD. Nonverbal expressions of affection were commonly described as the primary response to a child’s pain, as one parent explained:
‘The first thing [child] needs is just comfort, is somebody there. And then we discuss the levels of pain.’

(P12, Father of daughter, 12, HbSC).

Another parent describes her loss for words but instinctual comforting response: “I just hold her, I just don’t know what else to say” (Mother of daughter 7, HbSS).

Reports of these simple, but crucial, nonverbal responses indicate the importance of comfort over words when supporting their child during a crisis.

Communication of reassuring and compassionate messages was evident in parents’ descriptions of communication. These subliminal messages appeared to be aimed at preventing their child from experiencing guilt or isolation in response to learning about their condition. The following two excepts illustrate two key messages in turn: “It’s not your fault” and “You are not alone”, both of which were original codes in the data:

‘I remember the first thing he said “…well, but why me?... Did I do something wrong?” and I said “No you didn’t do anything wrong, it’s just, it’s a one in four chance, and unfortunately you got it, you know… it’s not your fault. It’s not your fault.’

(P10, Mother of son, 13, HbSS)

‘Just to explain there is not only you [child]”. You must know there’s some people with this, it’s not only, you [child] have pain, there is some children that also has different kind of blood disorders just to, to... because he said “Why me?”’

(P9, Father of son, 10, HbSbThal)
Whilst many had described efforts to make their child aware of their difference from others and potential limitations in life, some parents also expressed a determination for their child to feel hopeful about living with SCD and spoke of conversations that had taken place in which they had encouraged their child to live and aspire to a normal life; for example, one parent said: “I just encourage her. I say “You don’t let it define you”... “Don’t let it stop you”. [I] just encourage her.” (P1, Mother of daughter, 10, HbSS). Parents were therefore found to be making ongoing efforts to reframe potentially negative aspects of their child’s condition. Another example comes from a father of a 7-year-old girl with HbSS, who described efforts to reframe her disappointment about missing out on school trips, such as camping:

“You try to compensate. “You know, it’s not as good as a holiday and being in a nice hotel room and [laughs] getting the food served. So that’s different. You know “you won’t sleep very well [laughing] there”…”

(P2, Father of daughter, 7, HbSS)

Theme 3: Avoiding communication: “Don’t dwell on the negative”

Although parents spoke of having communicated openly about SCD, many also described having faced challenges that led them to avoid discussing SCD with their child. Avoidance was revealed in parents’ descriptions of ambiguity in their responses to child questioning. Many spoke about how they avoided the first discussion about SCD for some time despite their child’s evident curiosity about ‘why’ they have the condition, for example by responding:
‘That’s just how you are. Just how God made you.’

(P10, Mother of daughter, 10, HbSS)

First conversations were also triggered by questions about medication, which parents avoided answering directly:

‘Why is [my] sister not taking it?’ I say: ‘Because she don’t need it. It’s only you and your brother that need it.’

(P4, Mother of son, 10, HbSbThal).

Parents acknowledged, however, that this approach was not sustainable, recognising that they would eventually need to communicate more openly:

‘It’s difficult, it’s difficult. At times I don’t want to address the [SCD] topic at all but she will keep persisting. Even if I ignore it today I know she is going to ask me again.’

(P5, Mother of daughter, 10, HbSS)

Protection

Often avoidant communication resulted from parents’ efforts to protect their child from potentially distressing information. Some parents avoided informing young children about serious complications, including death, or negative accounts of the disease, reasons which P9 (Father of son,10, HbSbThal) explains:
‘I felt he was too young to understand or I might frighten him too much…we try to encourage him with positive things.’

Death was not the only avoided topic. Bone marrow transplant\(^3\), a potential cure for SCD was commonly avoided so as not to offer false hope to a child when the likelihood of a match was uncertain. One parent reflected, retrospectively, of their regret at not avoiding the topic:

‘Sometimes there is talk of having a bone marrow transplant. It was under discussion some time ago. She was involved in it she was… she got excited. So we get a sense she were [sic] a bit disappointed. So that was something we thought maybe we shouldn’t, you know, have got her involved in.’

(P2, Father of daughter, 7, HbSS)

A child’s age and capacity to understand was a commonly cited reason to avoid discussion about inheritance. Parental attitudes towards romantic relationships also impacted on their decisions about communicating:

‘We have not talked about [inheritance] because she’s still young she’s not thinking about marriage… we have kept it out because we are Christian … we don’t allow any sexual behaviour before marriage. When she is approaching that, we will talk about it. Fifteen, sixteen years, seventeen.’

(P12, Father of daughter, 12, HbSS)

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\(^3\) Also known as stem cell transplantation, a procedure that involves replacing bone marrow with marrow from a donor, resulting in normal haemoglobin production (Hsieh, Fitzhug & Weitzel et al., 2014)
Interestingly, the same parent spoke of avoiding ‘future talk’ altogether, in the hope that a cure would render present communication unnecessary:

‘We know everything will be alright… one day it will be a cure for the disease… we believe before she reach [sic] that time, she will be alright, that’s what we are hoping, that maybe we will not even need to talk to her because she will be well.’

Protecting their child from emotional distress was not the only motivation for avoiding discussion about SCD. Some parents described the emotional impact of talking about the negative aspects of SCD with their child. Several mothers, became upset during the interviews when talking about their child’s condition. One spoke of how this related to avoidance:

‘[Tearful] It’s hard for me to tell her that it’s going to be a…condition that she is going to be taking [medication] for the rest of her life. So, at times, because I don’t want her to see me crying, er, I don’t want to proceed so I try to cover it up with some other topic. So avoiding telling her that. It’s better I think.’

(P5, Mother of daughter, 12, HbSS)

Avoidant parents also expressed a desire for others to communicate with their child about SCD on their behalf:
‘So we have to make sure he is aware of a lot it. So if the [Sickle Cell Service] want to speak with him, advise him, educate him more about it, even to the mother, even to me because … I don’t know anything about it.’

(P11, Father of son, aged 7, HbSS)

Parental efforts to avoid communication appeared to be aimed at protecting both the parent and child from emotional distress. Interestingly, this approach appeared to be expressed by two fathers who were found to avoid discussion due to assumptions that the child was coping for example: “I think she is doing fine” (P2, Father of daughter, 7, HbSS), or by using strategies to distract a child from negative emotions:

‘So it’s recognising when she’s sad…and when she’s feeling sad… counteracting that… like at the moment she’s doing school work, so that’s distraction from being sad and focussing the “woe is me” and all that kind of stuff, and feeling down.’

(P8, Father of daughter, 11, HbSC)

Although this may signify an attempt to foster resilience, the approach risks the counteractive effect of dismissing the child’s emotional distress and conveying a message to the child that negative emotional expression is an unhealthy way of coping.
The Impact of Stigma

Stigma about SCD was raised by several parents who considered it as an ongoing issue in black and minority ethnic communities. A small number felt it had impacted on the frequency of communication about SCD within and outside of the family.

‘Even, you might be in the same house, you might have [extended family] living with you and those people don’t know because you don’t want them to know. You don’t want them to know because there’s a stigma to it, so it’s not something [parents] sit down and talk about - they don’t.’

(P1, Mother of daughter, 10, HbSS)

In response to the perception of stigma, communication was used to monitor outward disclosure about the child’s condition. The parent below describes the efforts she had made to ensure the condition had been kept hidden from peers and consequently the wider community:

‘He would say Mummy when [SCD nurse] came to school today my friends were asking me ‘Why is she here?’ ‘What has she come to do?’ Then I asked him what did you tell them? He said, I don’t know mummy, I just tell them that’s he has just come to check on me and my brother. I said “Okay that’s fine”…’

(P4, Mother of son, 10, HbSbThal)
The affirmation of her child’s response is reflective of her efforts to keep her son’s condition hidden. When asked how her children had learnt not to speak to others about their SCD, she responded:

‘I didn’t even say anything like ‘Don’t discuss it, don’t tell any of your friends’… Maybe because they see that I don’t discuss it with anyone.’

Children may therefore adopt avoidant communication patterns based on observations of what is ‘not said’ by parents.

Theme 4: Communication outcomes

The final theme encompasses the key outcomes of communication, as described by parents. Communication was described as impacting on a child’s knowledge about their condition, their role in managing SCD and their relationship with their parent.

Knowledge

Open communication, based on the principle of coaching, appeared to have led to improved knowledge about SCD which increased overtime. Knowledge also appeared to be an outcome of parents’ efforts to sensitively tailor communication to their child’s developmental capacity. Some parents had used communication aids, such as books, leaflets or online resources, to complement their communication in an attempt to transfer accurate information. Over time knowledge had become shared, with parents describing the development of mutual goals in promoting health and preventing crises.
Regarding their own levels of knowledge, a small number of parents expressed a lack of knowledge which was expressed explicitly: for example, “I know nothing about it” (P12, Father of son, 7, HbSS) and implicitly: “So I was thinking, is that part of the symptoms from the sickle, is it sickle beta, or something like that?” (P4, Mother of son, 7, HbSbThal).

**Role**

Children were described as becoming increasingly independent over time in managing in their condition, an outcome that parents believed had happened in response to their open and shared communication. Parents also spoke about communication leading to proactive and independent efforts by children to self-manage:

> ‘It’s changed in the form of, previously we had to remind her of what she, how she is to be dressed, … medication, in terms of [drinking fluids], she is more proactive on those terms. So even when you are not there she wants to do certain things that is needed.’

(P1, Mother of daughter, 10, HbSS)

Parent and child roles related to SCD management thus became shared over time as a result of parental communication about the condition. Management was described as being increasingly child led, with parents stepping back in managing their child’s health as their child gained knowledge, confidence and independent control of their condition.
**Relationships**

Positive relationships between parents and children were also described as an outcome of open and proactive parent-child communication. Within their relationships, parents described communication that resulted in children safely expressing their physical and emotional needs. Children were often described as expressing frustration during or after conversations about SCD, often in relation to missing out on activities with their peers or taking medication. This frustration was often validated by the parent and potential conflict was resolved through open communication. In contrast, one parent described how her relationship with her son had been affected by her own tendency to avoid talking, suggesting herself that she had passed her denial onto him. She spoke of her son’s resulting lack of acceptance, indicated through his expressions of frustration:

‘[Son says] “I have sickle it’s your fault”… “You should not have had me in the first place” … “If you had not married my dad in the first place I wouldn’t have had sickle cell”…’

(P10, Mother of son, 14, HbSS).

Another parent (P7, Mother of daughter, 10, HbSS) spoke about friction during conversations about self-management which had led to breakdowns in the relationship for “…days at a time”. She went on to explain how this had improved but was still a feature in their relationship: “…that’s when she was younger, now it’s like a couple of hours and she’s fine.”
**Discussion**

The aim of this research was to better understand the experiences of parents in communicating about sickle cell disorders (HbSS, HbSC & HbSbThal) and from this, make recommendations for addressing their support needs. Inductive thematic analysis enabled an open exploration of participant experience, enabling parents the opportunity to discuss the most meaningful elements of communication to them. In their interviews, parents described ways in which communication was initiated, approached and managed. The analysis offered insights into open and proactive communication patterns and also revealed the challenges parents face in communicating about the complications and limitations associated with SCD. Avoiding these topics posed risks to parent and child knowledge, their child’s capacity to take on an independent role and the quality of their relationship.

Parental attunement to a child’s physical and psychological wellbeing was found to be key in facilitating open communication about SCD. Close parental observation has been described as a common phenomenon in middle childhood, a time when emotional availability replaces physical proximity as a child’s main attachment need (Kerns, Aspelmeier, Gentzler & Grabill, 2001). Being open to communication and responsive to a child’s physical and emotional needs is a vital part of this process and is facilitative of secure attachments (Bowlby, 1987; Oppenhein & Waters, 1995). The physical effects and treatments associated with chronic illness, however, can be disruptive of attachment processes (Ødegård, 2005). Genetic conditions, such as SCD, may present additional challenges related to the hereditary basis of the disorder and the cultural stigma attached to it, which warrants further research. Understanding attachment patterns and how they influence parent-child communication in SCD may be of value in informing appropriate interventions aimed at enhancing adjustment.
A child’s socialisation to their condition was a common goal in parents’ explanatory communication with children. In providing physiological explanations of SCD, they aimed to motivate children to understand and manage their symptoms. They also provided reassurance in response to pain or emotional distress. In some cases, however, parents described more avoidant coping styles, which appeared to be based around their own fears and beliefs about the condition. Parents’ descriptions suggested that some children had developed maladaptive coping styles in response to a history of avoidant communication, which is supported by previous evidence of congruence in parent and child coping styles in SCD (Hildenbrand, Barakat, Alderfer & Marsac, 2015; Kliewer, Fearnow & Miller, 1996).

Previous qualitative research has identified that children with SCD desire open and honest communication from parents about their condition (Metcalfe, Plumridge, Shanks & Gill, 2008; Middleton et al., In Preparation). The present study provides evidence of parental motivation to meet this need. Honest communication that provides children with realistic expectations about their future is a factor that may promote quality of life in children with chronic illness, including SCD (Constantinou et al., 2015). By explaining and tailoring information about SCD to their children, parents described ‘coaching’ them in an open manner about the practicalities and limitations associated with their condition. However, whilst openness was commonly described, avoidant approaches were also evident, a finding that can be understood by Roth and Cohen’s (1986) Approach-Avoidance Model of Stress and Coping. This model has been used previously to understand general aspect of parent and child coping in SCD (Hildenbrand et al., 2015); however, the present study provides evidence of its potential applicability to communication patterns specifically. Within the model, ‘approaching’ is defined as efforts to direct
behaviour towards the management of a stressor, whereas avoidance includes people’s efforts to direct attention away (Roth & Cohen, 1986), both of which can be adaptive, depending on the stressor. ‘Approaching’ can be adaptive when dealing with controllable stressors (Manne, Bakeman & Jacobson, et al., 1993; Sorgen & Manne, 2002), such as medication and preventative behaviours, whilst avoidance may help manage time limited stressors or uncontrollable situations (Suls & Fletcher, 1985), such as an uncertain prognosis and painful crises. Aspects of parental avoidance observed in this study are thus not necessarily negative and can form adaptive ways of coping with uncontrollable and time limited situations, of which SCD presents many. A need therefore exists to identify maladaptive parental communication styles and to support them to tailor their communication in an adaptive way.

Regarding their support needs, parents spoke of requiring continuous and more detailed information to facilitate better communication with their child. A practical support system that includes information provision and instrumental help may be beneficial for and valued by parents. Many spoke of wanting others (i.e. healthcare professionals) to support them by communicating on their behalf, which has been found elsewhere in carrier studies (Ulph, Leong, Glazebrook & Townsend, 2010; Ulph et al., 2011). The present study suggests this desire may reflect parental avoidance and that healthcare professionals should explore parents’ underlying needs when making decisions about communication on their behalf. In addition to parents’ self-proclaimed lack of knowledge, their reluctance to communicate about certain issues may be reflective of difficulties with confidence and emotional resilience when tackling difficult issues such as a child’s prognosis and future with SCD. Indeed, interview data from several parents revealed the emotional impact of
having these discussions and a small number of parents expressed the need for emotional support from healthcare professionals with the more general challenge of parenting a chronically ill child. Overall, these findings are in line with the three dimensional definition of support outlined by Kirk & Glendinning (2002) with parents desiring both practical and emotional support from their child’s healthcare team. Provision of this style of support is therefore recommended and has potential to help parents develop realistic expectations of their child’s prognosis and improve the quality of parent-child interactions (Dinnebeil, 1999; Mahoney & Kaiser, 1999). There is growing evidence supporting the positive impact of parenting interventions, such as the triple p positive parenting programme which has recently been adapted for parent of children living with long term conditions (Morawska, Calam & Fraser, 2015). Parenting programmes include a number of key elements, including increasing positive interaction between parent and child as well as developing skills in communicating about emotions (Kaminski et al., 2008). Genetic conditions, such as SCD, have not yet been included in randomised controlled trials; however it is possible that such interventions could be adapted to address the support needs of the SCD parent population and further research is therefore indicated to explore this.

**Limitations**

The study was not without limitations. Firstly, the majority of parents in the study described proactive open communication and whilst representativeness was not a priority due to the in-depth qualitative nature of the study, it is important to acknowledge the potential impact of selection bias on study findings. That is, it is likely that those who communicated openly and were less avoidant were more likely to have participated in the study. Avoidant approaches, whilst apparent, may
therefore be underrepresented. Conclusions can thus only be provided on the nature of avoidance as a phenomenon and not of its prevalence.

Further consideration of the sample characteristics reveals that several of those who took part were well educated, many being graduates from higher education, reflecting a further limitation of selection bias. The potential impact of this on the transferability of the findings is also acknowledged. However, it is not considered an overall limitation due to the scope of the research questions and the underlying contextualist approach, within which findings are interpreted according to the local rather than global context.

Finally, whilst the aim of this study was to understand parental experience, implications for child adjustment and coping became a standalone theme but was derived solely from parent description. Thus, the true responses of children as well as their beliefs related to SCD cannot be inferred. Previous studies have indicated a need for more child based qualitative studies in SCD due to known inaccuracies in parental reports of child distress (Wade, Wilfond & McBride, 2010; Wilfond & Ross, 2009; Noke, Wearden, Peters & Ulph, 2014). The present findings provide additional evidence of the need to explore children’s own perceptions in order to more fully understand the influence and impact of parent and professional communication about SCD.
Conclusion

The findings of this study suggest that parental communication patterns in SCD may offer a useful way of understanding barriers and facilitators to a child’s adaption to the condition. Whilst parents displayed resilience in the face of difficulty and acted in ways to coach their child about SCD, avoidant communication styles were a common response to challenge. For some, this indicated difficulties with acceptance, whilst others described it as an adaptive response to suffering. Parents described communication styles that differed with those reported by and about healthcare professionals, who have been found to communicate more accurately and openly than parents (Gallo, Angst, Knafl, Twomey & Hadley, 2010; Middleton et al., In Preparation). Prospective studies of the development of health and illness beliefs in response to parent and professional communication patterns in SCD would inform interventions aimed at helping parents communicate openly and consistently with that of healthcare professionals. Providing parents with these skills offers to empower children with information that increases their understanding and ability to live well with SCD as children and into adulthood.

Declaration of Conflicting Interests

The authors declare that there is no conflict of interest.
References


A critical appraisal of the meta-synthesis and empirical study

Word Count: 4577 (Excluding references)
Overview
The present paper critically evaluates the meta-synthesis and empirical study described in this thesis. Critical reflections on various stages of the research process are presented, from planning and implementation to the analysis and interpretation of both studies. The strengths and limitations of each are discussed with implications for research and clinical practice outlined. The paper ends with some personal reflections about the research process in the context of the trainee’s learning and professional development.

Paper 1. The Meta-synthesis
Choosing a topic and method

Having decided to embark on an exploration of parent experiences of parent-child communication in sickle cell disease (SCD) for the empirical study, I was keen to conduct a literature review that was relevant to this topic but that provided greater insight into the child experience of communication. I hoped that together, the findings would enhance the knowledge base of parent and child communication in SCD and provide future directions for research and service development. Initial literature searches revealed a number of qualitative investigations of parent-child communication and of children’s communication with other groups. Thus, whilst the empirical area of enquiry related to parent-child communication, there appeared a need for considering communication between the child and both parent and non-parent groups. This topic was therefore chosen as the basis of the review.

Due to the predominance of qualitative studies identified in the exploratory searches, a meta-synthesis, informed by principles of meta-ethnography (Noblit &
Hare, 1988), was agreed as a suitable method following a discussion in supervision. The aim of the synthesis was to explore the impact that different avenues of communication had on the child, a topic so far unexplored in the literature. Within the studies initially identified, it became apparent that SCD populations are rarely studied as a group in their own right. Instead they tend to sit within a broad sample of genetic conditions, producing findings that are not necessarily reflective of people with SCD. A meta-synthesis of the studies was agreed to be advantageous in this sense, in that it permitted the extraction of quotations and findings relevant only to SCD, thereby facilitating the production of novel interpretations and more substantial insights into SCD than the studies alone produced (Finfgeld, 2003).

Conducting a meta-synthesis

Systematic approaches for the synthesis of quantitative studies are widely accepted and frequently utilised to collate and critically appraise existing research and interventions (Moher, Liber ate, Tetzlaff, & Altman, 2009; Mulrow, 1994). Increased publication of qualitative research has produced a need for a similar systematic methodology for accumulating and synthesising qualitative studies so that they can be reliably used to inform policy and practice (Walsh & Downe, 2006). According to Strike and Posner (1983), the goal of qualitative synthesis goes beyond that of narrative and systematic literature reviews to one of conceptual innovation and therefore requires a distinct approach. Noblit and Hare (1988) were among the first to advocate the utility of synthesising qualitative literature, albeit focussing exclusively on ethnographic research findings. An argument was later made for the inclusion of various methodologies within qualitative syntheses, given the paradigmatic notions of knowledge as constructed and truths as multiple (Walsh &
Downe, 2005). Noblit and Hare’s overall principles have however been retained and applied in efforts to synthesise qualitative research (Sandelowski, Docherty & Emden, 1997) and a number of attempts have been made to formalise the approach (Britten, Campbell, Pope, Donovan & Morgan, 2002, Thomas & Harden, 2008). The present study drew from approaches to meta-synthesis that were developed in maternity care research (Walsh & Downe, 2005; Downe, 2008) and utilised principles of meta-ethnography to produce new interpretations of communication experiences from the child’s perspective.

**Searching the Literature**

The development of research questions is a critical step that precedes effective literature searching (O’Connor, Green & Higgins, 2008). Cooke, Smith and Booth (2012) have developed a tool to assist researchers in developing qualitative research questions and effectively searching qualitative and mixed methods research. The ‘SPIDER’ tool provides a framework within which researchers are prompted to define the sample (S), phenomenon under investigation (PI), design (D), evaluative construct (e.g., views, attitudes) (E) and ‘research-type’ under examination (e.g. qualitative, quantitative and mixed-methods) (R). Use of this tool informed the research questions and facilitated a robust search. In parallel with the systematic searching stage conducted within systematic reviews, the goal was of retrieving recent (2005 - present) and relevant studies in the field (Barroso, Gollop, Sandelowski, Meynell, Pearce & Collins, 2003). Electronic searches were augmented with traditional reviewing methods, such as reference list screening and hand searching of journals, a process referred to elsewhere as ‘back-chaining’ (Downe, 2008). The application of search limits made the process more manageable.
in terms of screening the output however, despite this it still involved a significant investment of time to ensure inclusivity was maximised and screening was completed accurately. In order to ensure the review would be as up to date as possible at the time of submission, searches were repeated in August 2016 with citations arranged in date order so as to identify newly published literature. Surprisingly, no additional papers were identified for inclusion, which is reflective of the sparsity of published literature on communication in SCD.

Following careful screening, a total of 9 studies were included in the meta-synthesis. In comparison to systematic reviews, this may appear low but is quite typical of numbers included in meta-syntheses (Walsh & Downe, 2005). There has been a lack of discussion on the appropriate number of studies but instead an agreement that researchers should focus on ensuring that studies suitably address the research questions and scope of the synthesis. Thus, the aim of the presently described synthesis, as in all qualitative research, was not of representativeness but of providing evidence from which to interpret meaning (Baker, Edwards & Doidge 2012).

*Quality appraisal*

Clinicians and healthcare service managers are increasingly interested in utilising qualitative findings to improve healthcare services (Newman, Thompson & Roberts, 2006). Qualitative researchers are therefore challenged to produce quality research, which is suitable for forming a cumulative evidence base from which to inform practice (Murphy, Dingwall, Greatbatch, Parker & Watson, 1998). Debates of quality in qualitative research have typically centred around the historic struggle with positivist notions of ‘good’ research, often based on a search for ‘truth’ (Walsh...
& Downe, 2006). Adopting a subjectivist epistemology, qualitative research deprioritises the search for one truth and seeks to understand the complexities of human experience so as to create multiple interpretations of the data (Walsh & Downe, 2006). Healthcare commissioners, however, require some level of ‘truth’ to fund services in a safe, fair and reliable way. The extreme interpretivist approach therefore risks being deemed incompatible with the need for evidence based practice in healthcare (Murphy et al., 1998). Nevertheless, human understandings of experience are invaluable in healthcare evaluation (Walsh & Downe, 2006) and quality appraisals offer to enhance the credibility of qualitative research in healthcare (Thomas & Harden, 2008).

With this in mind, a quality checklist of criteria, developed by Walsh and Downe (2006), was used to appraise each study (Appendix 2). Each paper was assessed against the criteria, which included ‘Scope and Purpose’, ‘Appropriateness of Study Design’, ‘Sampling strategy’, ‘Analysis’, ‘Interpretation’, ‘Ethical Dimensions’ ‘Reflexivity’ and ‘Relevance and Transferability’. Thorough assessment of each study, against the criteria, informed the grading of studies according to four categories (see Box 1, below).

**Box 1. Grading system for qualitative study quality**

**Key to quality rating**

- **A** - No or few flaws, high credibility, transferability, dependability & confirmability of the study.

- **B** - Some flaws unlikely to affect credibility, transferability, dependability & confirmability of the study.

- **C** - Some flaws that may affect credibility, transferability, dependability & confirmability of the study.

- **D** - Significant flaws likely to affect credibility, transferability, dependability & confirmability of the study.

*(Downe, 2008)*
**Reflections conducting a meta-synthesis**

Following the selection of studies for inclusion, the next stage involved becoming immersed in the data by reading and re-reading each paper several times. This process was continued until I felt I had gained sufficient familiarity with the studies to notice patterns and compare common themes and findings. A significant amount of time was invested to facilitate this, a process that enabled the production of a coherent and valid representation of the collective findings.

Having chosen to use qualitative methods in both the literature review and empirical study, I made sure that papers one and two were analysed at distinct time points, that is, the thematic analysis of the empirical paper did not begin until the synthesis and writing up of paper one was completed. This was done purposefully in order to minimise confusion between the two analyses, which were similar in both topic and methodology.

**Limitations and Implications**

As stated in the original paper, the meta-synthesis included several studies that had reported different findings from the same sample. That is, Gallo, Angst, Knafl, Headley and Smith (2005) and Gallo, Angst and Knafl (2009) published findings on a genetic conditions sample that included 29 parents of children with sickle cell disease and Metcalfe, Plumridge, Coad, Shanks and Gill (2011), Plumridge, Metcalfe, Coad and Gill (2011a) and Plumridge, Metcalfe, Coad and Gill (2011b) all published findings on the same genetic conditions sample which included 33 families of children affected by sickle cell disease. The advantages and disadvantages of including these homogenous samples is acknowledged in the meta-synthesis paper and formed an early discussion in supervision. The decision to
include the entire group of papers was based on the lack of other research and the fact that each paper offered to present unique findings and novel interpretations related to SCD (Finfgeld, 2003).

It has also been considered that the review could have been expanded by including quantitative studies to form a mixed methods review. This was considered early on; however, the few quantitative studies that were identified were deemed not relevant to the research questions and therefore not compatible with the scope of the review.

Finally, whilst the meta-synthesis aimed to understand communication from the perspective of the child, this research question was only addressed through my own interpretations about children’s thoughts or feelings in response to the communication they had received. This critique is also, to some extent, relevant to the findings of the empirical study, which like paper one, implied a need for the voice of children in qualitative research in SCD rather than solely parent proxies. Shifts in the participatory rights of children following emphasis by the United Nations Convention on the Rights of the Child (UNCRC, 2002) and an interest in children as “...subjects and active agents experiencing and shaping their own lives” (Hill, 2006 pp.72), have meant that children are increasingly included in research, policy planning and service level consultation (Hill, Davis, Prout & Tisdall, 2004). Increased qualitative research including children with SCD is therefore both appropriate and necessary in order to understand their perspectives and design services which meet their needs.

**Appraisal of Paper 2: The empirical study**

The proposal for the empirical study emerged from my interests in clinical health psychology and following my experience of working within a sickle cell and
thalassaemia service as an Assistant Psychologist. In this role I had become aware of the psychological impact of sickle cell disease as well as the potential problems of non-adherence, which was particularly evident in poor adult outpatient attendance. Indeed, research suggests that adults with SCD often stay away from hospital despite experiencing problems coping in the community (Maxwell, Streetly, & Bevan, 1999). With this in mind, I was interested in exploring factors that predisposed some people with SCD to maladaptive styles of coping. Theories of illness beliefs suggest that they are developed early on in life through social learning and communication (DiMatteo, Haskard & Williams, 2007). The design of the empirical study was therefore based on my prior interests and experience, together with indications from research evidence and theory.

**Research questions and choice of method**

Having identified gaps in the literature on family communication in SCD, research questions were decided on through discussion in supervision. Questions were kept broad and exploratory to facilitate rich qualitative enquiry and to capture new understandings of the experiences of a poorly researched group. Further decisions were made in supervision about data collection methods and plans for the analytic approach. Thematic analysis was chosen as a suitable approach for producing rich findings without fixed prescriptions about data collection or epistemological framework (Clarke & Braun, 2014). Thematic analysis is an increasingly popular method in social science research, despite having only recently been recognised as a distinctive method with a clear set of procedures (Braun & Clark, 2006). The methodological and epistemological flexibility offered in this analytic approach were deemed suitable for the broad research questions and scope of the study.
By adopting an epistemological stance of contextualism, the study aimed to understand parental accounts of communication within the context of factors that may influence these accounts, for example, socio-political issues; personal views and experiences. As a condition affecting black and minority ethnic (BME) groups, SCD is surrounded by issues of race and culture, including prejudice and illness related stigma from both within and outside of the BME community (Jenerette, Funk & Murdaugh, 2005). These embedded socio-economic issues were considered throughout the empirical study and research questions were explored broadly, as was intended, by interpreting participant accounts in context.

Recruitment

Qualitative studies typically recruit lower numbers of participants, however, the decision about sample size is not necessarily straightforward (Sandelowski, 1995). Such decisions are influenced by the scope and purpose of the study, the quality of data collected and the point at which data saturation\(^4\) is reached (Morse, 2000). Thus, a number of factors can influence decisions about sample size at various stages of the research process. In the present study, the initial proposed sample was of 20 participants; however, a smaller sample of 12 resulted. Interviews were reviewed mid-way through the recruitment phase and two observations were made following discussion in qualitative supervision. Firstly, uptake had been slower than we had envisaged with 6 participants recruited, which was just over a quarter of the original target of 20. Secondly, despite small in number, the interviews had provided good quality data that were nearing saturation. The focus for the remaining phase of recruitment therefore changed to gaining greater diversity in the sample and a shift to

\(^4\) That is, the point at which further analysis is no longer revealing novel concepts and themes.
more purposive sampling resulted. This strategy offered to include varying
perspectives and produce broad and rich data that generated deep insight into
participant experience (Patton, 2002). Recruitment was targeted at people of varying
socioeconomic backgrounds, different sickle cell diagnoses (i.e. HbSS, HbSC,
HbSbThal), fathers, mothers and parents of children of various ages. These wide
ranging characteristics offered to provide rich understandings of different lived
experiences of communicating about SCD with an affected child.

**Conducting the interviews**

With support from the field supervisor, a member of the local sickle cell parent
support group was approached to pilot the interview schedule. The use and
importance of piloting in qualitative research is widely acknowledged (Breakwell,
2006; Sampson, 2004). Piloting the schedule was a key stage in testing the flow of
the schedule and helped ensure questions were written in a comprehensible style.
The process also welcomed feedback from the ‘pilot participant’, which was
incorporated into the final schedule (Appendix 9). For example, the pilot participant
suggested adding questions about ‘not talking’, that is, “What do you not talk
about?” This question facilitated discussion about commonly described ‘difficult
discussions’ and the topic of avoidance, which became a key theme with critical
implications for research and practice.

Once the interviews were underway, I noticed becoming more comfortable
with the schedule. I became increasingly familiar with the questions and arising
issues, which arguably helped put participants at ease and build a better rapport.
Furthermore, many interviews were conducted in participant homes, which rendered
the process less formal and helped create a sense of safety for them to share their experiences.

_The evolving nature of analysis_

The analysis took on several forms throughout the various stages of the process. Initial thoughts and ideas about recurring and important findings began to emerge early on, during the interview and transcription stages. Completing the interviews and the majority of transcription myself was advantageous to the analysis. My own deep immersion in the interviews and transcripts provided a sense of really ‘knowing’ the data. That is, I was familiar with the multitude of stories and experiences shared but also experienced a felt awareness of nuances within the data having bared witness to parents’ emotive accounts.

Following transcription, coding was completed for the entire data set. Whilst initial coding remained at the basic level, the stage that followed took a more latent approach by analysing the data from a contextualist position. A number of computer software packages have been designed for qualitative data analysis to enable the complex organisation and retrieval of data and improve rigour (Pope, Zeibland & Mays, 2000). For these reasons transcripts were uploaded and coded on the software package Nvivo (Version 10). The process of coding was thus straightforward and reliable. That is, codes were not lost, forgotten or confused with others but were reliably recorded. The software also aided the process of generating themes by collapsing or clustering codes that appear to share a unifying feature and reflected coherent and meaningful patterns (Braun, Clark & Terry, 2014). Scanning the initially very large number of codes revealed a multitude of possibilities for patterns and relationships. This became more manageable as codes were grouped, merged or
collapsed to represent similar or corresponding concepts. This process was continued, and led to the production four themes. Excerpts were chosen from coded data to capture and represent each theme clearly. These were written up in draft form, shared with the primary supervisor (FU), adapted again and written up, the latter of which marked the final stage of the analysis (Braun & Clarke, 2013). On reflection, acceptance of the changing ideas and frameworks required some discipline in not becoming overly attached to ideas and candidate themes. Supervision was particularly helpful for this by providing the opportunity to question and justify themes, provide alternative interpretations and ensure that the framework adhered to principles of Thematic Analysis.

*Inter-rater reliability*

Inter-rater reliability in qualitative research refers to the comparison of independently coded data to provide a level of agreement, and is often claimed to be suggestive of empirical quality (Mays & Pope, 1995). The necessity of independent ratings has however been subject to debate (Armstrong, Gosling, Weinman & Marteau, 1997). Arguments for and against using inter-rater checks sit within long-standing ontological debates around understandings of ‘reality’. Those against systematic comparisons of coding decisions have argued that it is unrealistic to expect second readers to gain identical insights from small samples of data (Morse, 1994). With this in mind, inter-rater reliability measures were decided against in the empirical study. Instead, the reliability and validity of the study were promoted through regular discussion in supervision including the sharing of emerging ideas and written reflections about the data. Regular face to face, email and phone contact with the primary thesis supervisor was maintained throughout the analysis phase and
minutes of meetings and discussions were shared with the research team. Opportunities were thus provided for others to comment on the integrity of emergent themes, whilst preserving the subjective interpretations developed from my own immersion in data collection and analysis.

**Maintaining reflexivity**

The influence of a researcher’s own views on the research process is a widely accepted phenomenon, known to influence even the most rigorous of inductive qualitative inquiries (Mays & Pope, 2000). Maintaining reflexivity requires one’s awareness and acknowledgment of the influence of personal values and experience on the research process, something that reflective journals can facilitate (Ortlipp, 2008). Monitoring and acknowledgement of reflexivity offer to enhance the credibility of qualitative research (Armstrong et al., 1997) and may offer to improve the quality of the process (Ortlipp, 2008). For these reasons, a reflective journal was kept throughout data collection and analysis. I also had the opportunity of debriefing with my supervisor after interviews, which, in addition to regular supervision helped me maintain a balanced view when making initial interpretations. The journal was particularly helpful in enabling me to record my observations during interviews and provided space to document powerful feelings that emerged during interviews. The following excerpt illustrates this and was one I held in mind during the analysis and write up to retain this focus:

*I’m feeling so moved by the interviews so far. By the single mum coping alone with a chronically ill child, and the crushing feelings of guilt some parents describe about their sense of responsibility for their child’s pain and*
prognosis. I’m also inspired by parents’ dedication and sacrifice - some having put life completely on hold to support their child and experiencing loss and regret as a result. Who is supporting them with this?

Regarding the impact of my own experience, my prior experience working with people affected by sickle cell disease and my current transition into parenthood were considered as shared qualities that may have influenced the research process. The acknowledgment of such shared qualities may have acted to enhance the credibility of the findings in that I was able to sit alongside participants when interpreting the data. My familiarity with sickle cell disease and my status as a parent were also qualities I felt I shared with participants and helped facilitate rapport. My status as a parent was also one that may have led me to feel more sympathetic towards parents and less blaming of behaviours, such as avoidance. It is possible that this encouraged me to rationalise their behaviour more than a non-parent would, for example, by focussing on ‘protection’ as a factor in parental avoidance.

Conversely, acknowledging the ‘distance’ between researcher and participants is an important consideration in reflexive discussion (Mays & Pope, 2000). As a White-British female who, despite being a parent, has a healthy child unaffected by genetic illness, our experiences are likely to have been distinct in many ways. Illnesses that affect my own child have been less serious, however my ability to access and navigate within and around the healthcare systems for my own child is likely to have been more manageable compared to that of the participants, many of whom were not NHS employees and had not been born in the UK. Additional differences in our experiences of culture, although mainly unspoken, may well have impacted on participants’ willingness to share their account with me. It
may also have caused me to misinterpret, or miss altogether, aspects of cultural significance, such as stigma, that were distinct from my own non-stigmatising perceptions of SCD. An additional phase of ‘member checking’ may have offered to address this issue. Within this strategy, a researcher’s interpretations are shared with participants to gain feedback on the accuracy of the interpretations and to address areas of miscommunication (Johnson, 1997). On reflection, the pilot participant may have been a suitable person to ask to complete this exercise.

Finally, the therapeutic skills I have developed during the ClinPsyD were undoubtedly helpful in building rapport with participants, as well as in creating a safe and containing space during the interviews. It was however important, and somewhat challenging at times, to maintain the boundaries of my role as a researcher. Whilst making efforts to ensure my questions adhered to the interview schedule, it was a challenge to withhold my reflections, interpretations or suggestions, which would have been shared in therapeutic discussions.

Clinical implications and future directions

Whilst the presence of psychological factors in SCD have been acknowledged in research and clinical practice (Anie, 2005; Sickle Cell Society, 2008), the way in which these factors develop and impact on later management of SCD have been poorly understood by healthcare professionals (Maxwell, Streetly, & Bevan, 1999). Improving understanding of parental influences on the development of health beliefs may therefore help professionals to support them to hold open and honest discussions that are consistent with those held in medical settings. Together, the findings of papers one and two suggest that parental avoidance is common which may affect children and young people’s coping and adjustment due to the different
messages they hear from different personal and professional groups. Parents of children with SCD would therefore benefit from support to ensure they feel able to communicate openly and effectively with their child in a way that is consistent with healthcare professionals. Data from the interviews suggest that parents are keen to receive such support and would be accepting were it to be offered. Healthcare professionals, such as sickle cell and thalassaemia specialist nurses, social workers and psychologists working in paediatric Sickle Cell and Thalassaemia Services, are well placed to identify and help those in need of support. Explorations focussing on children with SCD were also indicated. Of particular interest would be prospective investigations of the influence of child health and illness beliefs on future coping in SCD. Studies such as this may inform interventions aimed at preventing long term psychological morbidity in SCD.

**Personal reflections**

The ability to conceptualise, design and conduct independent, original research is a skill within the ‘Research’ core competency that doctoral trainees must demonstrate to qualify as a Clinical Psychologist (British Psychological Society, 2014). Additional skills in psychological assessment, formulation and intervention, as well as teaching and leadership are assessed academically and on clinical placements. The programme is therefore extremely varied and requires careful management of time and workload throughout. Nevertheless, maintaining momentum with the large scale research project (LSRP) through the peaks of troughs of the programme has been a challenge. Working with numerous competing demands, whilst striving to maintain a work-life balance, has been a useful learning experience that will no doubt be required in my career as a Clinical Psychologist. Despite the inherent challenges, the
LSRP has also offered a number of new learning opportunities including new knowledge and skills in meta-synthesis, the experience of using quality assessment tools for reviewing qualitative literature and further development of my knowledge and skills in qualitative analysis.

It is interesting to reflect that during my final year placement, I have been approached by several practitioners requesting advice or guidance about research processes, for example, writing protocols, research ethics procedures and data collection methods. These requests have often come from Clinical Psychologists, who claim that whilst they are keen to conduct empirical research, the demands of their clinical role render such work practically impossible. Instead, department research in this particular setting mainly consists of audit or service evaluation, which is often completed by Trainee Clinical Psychologists. This reflection is not intended to criticise, but to instead help me envisage the potential difficulties I may face in implementing research within my role as a Clinical Psychologist and consider ways to overcome them. My hopes for post qualification are to continue research activity in areas exploring psychological wellbeing in long term conditions and to gain employment within a Clinical Health Psychology Service.

Increased austerity measures on NHS resources have resulted in reductions in funding for numerous mental health and psychology services, including psychology in health settings. Existing services are therefore under threat and practitioners work within an environment of increasing demand. The need for evidence based research by the qualified, scientist-practitioners working within these areas is thus more important than ever. It is essential for clinicians to be measuring the impact and effectiveness of their work by collecting and publishing outcome data. Conducting applied research such as this is likely to result in convincing evidence for presenting
to NHS commissioners who fund psychology in health settings, and is a role I aspire to in entering the profession.
References


Appendix
Appendix 1: Contributor guidelines for Social Science and Medicine

**SOCIAL SCIENCE & MEDICINE**

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**DESCRIPTION**

*Social Science & Medicine* provides an international and interdisciplinary forum for the dissemination of *social science* research on *health*. We publish original research articles (both empirical and theoretical), reviews, position papers and commentaries on health issues, to inform current research, policy and practice in all areas of common interest to social scientists, health practitioners, and policy makers. The journal publishes material relevant to any aspect of health from a wide range of social science disciplines (anthropology, economics, epidemiology, geography, policy, psychology, and sociology), and material relevant to the social sciences from any of the professions concerned with physical and mental health, health care, *clinical practice*, and *health policy* and organization. We encourage material which is of general interest to an international readership.

The journal publishes the following types of contribution:

1) Peer-reviewed original research articles and critical or analytical reviews in any area of socialscience research relevant to health. These papers may be up to 8,000 words including abstract, tables, and references as well as the main text. Papers below this limit are preferred.

2) Peer-reviewed short reports of research findings on topical issues or published articles of between2000 and 4000 words.
3) Submitted or invited commentaries and responses debating, and published alongside, selected articles.

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AUTHOR INQUIRIES
Visit the Elsevier Support Center to find the answers you need. Here you will find everything from Frequently Asked Questions to ways to get in touch. You can also check the status of your submitted article or find out when your accepted article will be published.

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Appendix 2. Complete quality analysis

<table>
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<tr>
<th>Criteria</th>
<th>Essential</th>
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<th>3</th>
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<th>6</th>
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<tbody>
<tr>
<td>Scope and Purpose</td>
<td>Clear Statement of, and rationale for, research question/ aims/ purposes</td>
<td>***</td>
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<td>Study thoroughly contextualised with research intent</td>
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<tr>
<td>Design</td>
<td>Method/ design apparent and consistent with research intent</td>
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<td>Data collection strategy apparent and appropriate</td>
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<td>Sampling Strategy</td>
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<td>Analysis</td>
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<td>Clear audit trail given</td>
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<td>Data used to support interpretation</td>
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<td>Reflexivity</td>
<td>Researcher reflexivity demonstrated</td>
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<td>Ethical dimensions</td>
<td>Demonstration of sensitivity to ethical concerns</td>
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<td>Relevance and</td>
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<td>A</td>
<td>A</td>
<td>A</td>
<td>A</td>
<td>C</td>
</tr>
</tbody>
</table>

⁵ Quality rating key: A: no or few flaws, high credibility; B: some flaws unlikely to affect credibility; C: some flaws that may affect credibility; D: significant flaws likely to affect credibility of the study.
Appendix 3: Contributor guidelines for Qualitative Health Research

Author Guidelines: Qualitative Health Research (QHR)

1. Article Types
   1.1 What type of articles will QHR accept?

2. Editorial Policies
   2.1 Peer review policy
   2.2 Authorship
   2.3 Acknowledgements
   2.4 Funding
   2.5 Declaration of conflicting interests
   2.6 Research ethics and patient consent
   2.7 Clinical trials
   2.8 Reporting guidelines
   2.9 Data

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   3.1 Publication ethics
   3.2 Contributor’s publishing agreement
   3.3 Open access and author archiving
   3.4 Permissions

4. Preparing your Manuscript
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   4.2 Word processing formats
   4.3 Artwork, figures and other graphics
   4.4 Supplementary material
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   6.2 SAGE Production
   6.3 Access to your published article
   6.4 Online First publication
   6.5 Open Access and SAGE Choice

7. Additional Information
1. Article types

Each issue of QHR provides readers with a wealth of information — book reviews, commentaries on conceptual, theoretical, methodological and ethical issues pertaining to qualitative inquiry as well as articles covering research, theory and methods.

1.1 What types of articles will QHR accept?

Authors should make the initial decision regarding the fit of their article to the journal. Do not send query letters regarding article fit.

- Read the Mission Statement on main QHR webpage.
- Search the QHR journal for articles that address your topic. Do we publish in your area of expertise?
- Ask these questions: Does your article make a meaningful and strong contribution to qualitative health research literature? Is it original? Relevant? In depth? Insightful? Significant? Is it useful to reader and/or practitioner?
- Note the sections: General articles, critical reviews, articles addressing qualitative methods, commentaries on conceptual, theoretical, methodological, and ethical issues pertaining to qualitative inquiry.
- QHR accepts qualitative methods and qualitatively-driven mixed-methods, qualitative metaanalyses, and articles addressing all qualitative methods.
- QHR is a multi-disciplinary journal and accepts articles written from a variety of perspectives including: cross-cultural health, family medicine, health psychology, health social work, medical anthropology, medical sociology, nursing, pediatric health, physical education, public health, and rehabilitation.
- Articles in QHR provide an array of timely topics such as: experiencing illness, giving care, institutionalization, substance abuse, food, feeding and nutrition, living with disabilities, milestones and maturation, monitoring health, and children’s perspectives on health and illness.

Look Out for These Regular Special Features

Pearls, Pith and Provocation: This section fosters debate about significant issues, enhances communication of methodological advances and encourages the discussion of provocative ideas.
**Book Review Section:** *Qualitative Health Research* includes a book review section helping readers determine which publications will be most useful to them in practice, teaching and research.

**Mixed Methods:** This section includes qualitatively-driven mixed-methods research, and qualitative contributions to quantitative research.

**Advancing Qualitative Methods:** Qualitative inquiry that has used qualitative methods in an innovative way.

**Evidence of Practice:** Theoretical or empirical articles addressing research integration and the translation of qualitatively derived insights into clinical decision-making and health service policy planning.

**Ethics:** Quandaries or issues that are particular to qualitative inquiry are discussed.

**Teaching Matters:** Articles that promote and discuss issues related to the teaching of qualitative methods and methodology.

### 2. Editorial policies

#### 2.1 Peer review policy

QHR strongly endorses the value and importance of peer review in scholarly journals publishing. All articles submitted to the journal will be subject to comment and external review. All manuscripts are initially reviewed by the Editors and only those articles that meet the scientific and editorial standards of the journal, and fit within the aims and scope of the journal, will be sent for outside review.

QHR adheres to a rigorous double-blind reviewing policy in which the identity of both the reviewer and author are always concealed from both parties. Ensure your manuscript does not contain any author identifying information. Please refer to the editorial on blinding found in the Nov 2014 issue: [http://qhr.sagepub.com/content/24/11/1467.full](http://qhr.sagepub.com/content/24/11/1467.full).

QHR maintains a transparent review system, meaning that all reviews, once received, are then forwarded to the author(s) as well as to ALL reviewers.

Peer review takes an average of 6–8 weeks, depending on reviewer response.

#### 2.2 Authorship

Articles should only be submitted for consideration once all contributing authors give consent. Those submitting articles should carefully check that all those whose work contributed to the article are acknowledged as contributing authors.

The list of authors should include all those who can legitimately claim authorship.

This is all authors who:
(i) Made a substantial contribution to the concept and design, acquisition of data or analysis and interpretation of data,
(ii) Drafted the article or revised it critically for important intellectual content, (iii) Approved the version to be published.

Authors should meet the conditions of all of the points above. Each author should have participated sufficiently in the work to take public responsibility for appropriate portions of the content.

When a large, multicenter group has conducted the work, the group should identify the individuals who accept direct responsibility for the manuscript. These individuals should fully meet the criteria for authorship.

Acquisition of funding, collection of data, or general supervision of the research group alone does not constitute authorship, although all contributors who do not meet the criteria for authorship should be listed in the Acknowledgments section. Please refer to the International Committee of Medical Journal Editors (ICMJE) authorship guidelines for more information on authorship.

2.3 Acknowledgements
All contributors who do not meet the criteria for authorship should be listed in an Acknowledgements section. Examples of those who might be acknowledged include a person who provided purely technical help, or a department chair who provided only general support.

2.3.1 Writing assistance
Individuals who provided writing assistance, e.g., from a specialist communications company, do not qualify as authors and should only be included in the Acknowledgements section. Authors must disclose any writing assistance — including the individual’s name, company and level of input — and identify the entity that paid for this assistance.

It is not necessary to disclose use of language polishing services.

Please supply any personal acknowledgements separately from the main text to facilitate anonymous peer review.

2.4 Funding
QHR requires all authors to acknowledge their funding in a consistent fashion under a separate heading. Please visit the Funding Acknowledgements page to confirm the format of the acknowledgement text in the event of funding. Otherwise, state that: This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

2.5 Declaration of conflicting interests
It is the policy of QHR to require a declaration of conflicting interests from all authors enabling a statement to be carried within the paginated pages of all published articles. Please ensure that a “Declaration of Conflicting Interests”
statement is included at the end of your manuscript, after any acknowledgements and prior to the references. If no conflict exists, please state that “The Author(s) declare(s) that there is no conflict of interest.”

For guidance on conflict of interest statements, please see the ICMJE recommendations here.

2.6 Research ethics and patient consent
Medical research involving human subjects must be conducted according to the World Medical Association Declaration of Helsinki.

Submitted manuscripts should conform to the ICMJE Recommendations for the Conduct, Reporting, Editing, and Publication of Scholarly Work in Medical Journals, and all articles reporting animal and/or human studies must state in the methods section that the relevant Ethics Committee or Institutional Review Board provided (or waived) approval. Please ensure that you have provided the full name and institution of the review committee, in addition to the approval number.

For research articles, authors are also required to state in the methods section whether participants provided informed consent and whether the consent was written or verbal.

In terms of patient privacy, authors are required to follow the ICMJE Recommendations for the Protection of Research Participants. Patients have a right to privacy that should not be infringed without informed consent. Identifying information, including patients' names, initials, or hospital numbers, should not be published in written descriptions, photographs, and pedigrees unless the information is essential for scientific purposes and the patient (or parent or guardian) gives written informed consent for publication. Informed consent for this purpose requires that a patient who is identifiable be shown the manuscript to be published. Participant descriptors should not be listed individually. Because qualitative research is descriptive, it is recommended that participant quotations not be linked to identifiers in the manuscript.

2.7 Clinical trials
QHR conforms to the ICMJE requirement that clinical trials are registered in a WHO-approved public trials registry at or before the time of first patient enrolment as a condition of consideration for publication. The trial registry name and URL, and registration number must be included at the end of the abstract.

2.8 Reporting guidelines
The relevant EQUATOR Network reporting guidelines should be followed depending on the type of study. For example, all randomized controlled trials submitted for publication should include a completed Consolidated Standards of Reporting Trials (CONSORT) flow chart as a cited figure, and a completed CONSORT checklist as a supplementary file.
Other resources can be found at NLM’s Research Reporting Guidelines and Initiatives.

2.9 Data
SAGE acknowledges the importance of research data availability as an integral part of the research and verification process for academic journal articles.

QHR requests all authors submitting any primary data used in their research articles alongside their article submissions to be published in the online version of the journal, or provide detailed information in their articles on how the data can be obtained. This information should include links to third-party data repositories or detailed contact information for third-party data sources. Data available only on an author-maintained website will need to be loaded onto either the journal’s platform or a third-party platform to ensure continuing accessibility. Examples of data types include but are not limited to statistical data files, replication code, text files, audio files, images, videos, appendices, and additional charts and graphs necessary to understand the original research. [The editor(s) may consider limited embargoes on proprietary data.] The editor(s) [can/will] also grant exceptions for data that cannot legally or ethically be released. All data submitted should comply with Institutional or Ethical Review Board requirements and applicable government regulations. For further information, please contact the editorial office at vshannonqhr@gmail.com.

3. Publishing Policies

3.1 Publication ethics
SAGE is committed to upholding the integrity of the academic record. We encourage authors to refer to the Committee on Publication Ethics’ International Standards for Authors and view the Publication Ethics page on the SAGE Author Gateway.

3.1.1 Plagiarism
QHR and SAGE take issues of copyright infringement, plagiarism or other breaches of best practice in publication very seriously. We seek to protect the rights of our authors and we always investigate claims of plagiarism or misuse of articles published in the journal. Equally, we seek to protect the reputation of the journal against malpractice. Submitted articles may be checked using duplication-checking software. Where an article is found to have plagiarized other work, or included third-party copyright material without permission, or with insufficient acknowledgement, or where authorship of the article is contested, we reserve the right to take action including, but not limited to: publishing an erratum or corrigendum (correction); retracting the article (removing it from the journal); taking up the matter with the head of department or dean of the author’s institution and/or relevant academic bodies or societies; banning the author from publication in the journal or all SAGE journals, or appropriate legal action.

3.2 Contributor’s publishing agreement
Before publication, SAGE requires the author as the rights holder to sign a Journal
Contributor’s Publishing Agreement. SAGE’s Journal Contributor’s Publishing Agreement is an exclusive license agreement which means that the author retains copyright in the work but grants SAGE the sole and exclusive right and license to publish for the full legal term of copyright. Exceptions may exist where an assignment of copyright is required or preferred by a proprietor other than SAGE. In this case copyright in the work will be assigned from the author to the society. For more information please visit our Frequently Asked Questions on the SAGE Journal Author Gateway.

3.3 Open access and author archiving
QHR offers optional open access publishing via the SAGE Choice program. For more information please visit the SAGE Choice website. For information on funding body compliance, and depositing your article in repositories, please visit SAGE Publishing Policies on our Journal Author Gateway.

3.4 Permissions
Authors are responsible for obtaining permission from copyright holders for reproducing any illustrations, tables, figures or lengthy quotations previously published elsewhere. For further information including guidance on fair dealing for criticism and review, please visit our Frequently Asked Questions on the SAGE Journal Author Gateway.

4. Preparing your manuscript

4.1 Article Format (see previously published articles in QHR for style):

- **Title page:** Title should be succinct; list all authors and their affiliation; keywords. Please upload the title page separately from the main document.
- **Blinding:** Do not include any author identifying information in your manuscript, including author’s own citations. Do not include acknowledgements until your article is accepted and unblinded.
- **Abstract:** Unstructured, 150 words. This should be the first page of the main manuscript, and it should be on its own page.
- **Keywords:** Listed below the abstract, on the same page.
- **Length:** QHR does not have a word or page count limit. Manuscripts should be as tight as possible, preferably less than 30 pages including references. Longer manuscripts, if exceptional, will be considered.
- **Methods:** QHR readership is sophisticated; excessive details not required.
- **Ethics:** Include a statement of IRB approval and participant consent. Present demographics as a group, not listed as individuals. Do not link quotations to particular individuals unless essential (as in case studies) as this threatens anonymity.
- **Results:** Rich and descriptive; theoretical; linked to practice if possible.
- **Discussion:** Link your findings with research and theory in literature, including other geographical areas and quantitative research.
• **References:** APA format. Use pertinent references only. References should be on a separate page. [Visit this link for APA guidelines](#). • Use Endnote formatting, not Footnotes.

**Additional:**
• Please do not refer to your manuscript as a “paper;” you are submitting an “article.” □ The word “data” is plural.

### 4.2 Word processing formats
Preferred formats for the text and tables of your manuscripts are Word DOC or PDF. The text should be double-spaced throughout with a minimum of 1.25 inches for left- and right-hand margins and 2 inches at head and foot. Text should be standard font (i.e., Times New Roman) 10 or 12 point.

### 4.3 Artwork, figures and other graphics
• Figures: Should clarify text. Do not use shading inside boxes. Use only black font.
• Figures, tables, and other files should be uploaded separately from the main document. Indicate where the table/figure should be inserted within manuscript text (i.e., INSERT TABLE 1 HERE).
• Photographs: Should have permission to reprint and faces should be concealed using mosaic patches – unless permission has been given by the individual to use their identity. This permission must be forwarded to QHR’s Managing Editor.
• TIFF, JPED, or common picture formats accepted. The preferred format for graphs and line art is EPS.
• Resolution: Rasterized based files (i.e. with .tiff or .jpeg extension) require a resolution of at least **300 dpi** (dots per inch). Line art should be supplied with a minimum resolution of **800 dpi**.
• Dimension: Check that the artworks supplied match or exceed the dimensions of the journal. Images cannot be scaled up after origination.
• Figures supplied in color will appear in color online regardless of whether or not these illustrations are reproduced in color in the printed version. For specifically requested color reproduction in print, you will receive information regarding the costs from SAGE after receipt of your accepted article.

### 4.4 Supplementary material
This journal is able to host additional materials online (e.g., datasets, podcasts, videos, images, etc.) alongside the full-text of the article. These will be subjected to peer-review alongside the article.

Supplementary files will be uploaded as supplied. They will not be checked for accuracy, copyedited, typeset or proofread. The responsibility for scientific accuracy and file functionality remains with the author(s). SAGE will only publish supplementary material subject to full copyright clearance. This means that if the content of the file is not original to the author, then the author will be responsible for clearing all permissions prior to publication. The author will be required to
provide copies of permissions and details of the correct copyright acknowledgement.

4.5 Journal layout
In general, QHR adheres to the guidelines contained in the Publication Manual of the American Psychological Association [“APA”], 6th edition (ISBN 10:1-4338-0561-8, softcover; ISBN 10:1-4338-0559-6, hardcover; 10:1-4338-0562, spiral bound), with regard to manuscript preparation and formatting. These guidelines are referred to as the APA Publication Manual, or just APA. Additional help may be found online at http://www.apa.org/, or search the Internet for “APA format.”

4.6 Reference style
QHR adheres to the APA reference style. Click here to review the guidelines on APA to ensure your manuscript conforms to this reference style.

4.7 English language editing services
Articles must be professionally edited; this is the responsibility of the author. Authors seeking assistance with English language editing, translation, or figure and manuscript formatting to fit the journal’s specifications should consider using SAGE’s Language Services.

4.8 Review Criteria
Before submitting the manuscript, authors should have their manuscript pre-reviewed using the following QHR criteria:

<table>
<thead>
<tr>
<th>1. Importance of submission:</th>
<th>Does it make a meaningful and strong contribution to qualitative health research literature? Is it original? Relevant? In depth? Insightful? Significant? Is it useful to reader and/or practitioner?</th>
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<tbody>
<tr>
<td>2. Theoretical orientation and evaluation:</td>
<td>Is it theoretically clear and coherent? Is there logical progression throughout?</td>
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<tr>
<td>4. Ethical Concerns (Including IRB approval and consent):</td>
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<tr>
<td>5. Data analysis and findings:</td>
<td>Does the analysis of data reflect depth and coherence? In-depth descriptive and interpretive dimensions? Creative and insightful analysis? Linked with theory? Relevant to practice/discipline?</td>
</tr>
</tbody>
</table>
6. **Data analysis and findings**: Does the analysis of data reflect depth and coherence? In-depth descriptive and interpretive dimensions? Creative and insightful analysis? Linked with theory?

7. **Discussion**: Results linked to literature? Contribution of research clear? Relevant to practice/discipline?

8. **Manuscript style and format**: Please evaluate writing style: Length (as short as possible], organization, clarity, grammar, appropriate citations, etc.); presentation of diagrams/illustrations?

---

5. Submitting your manuscript

5.1 **How to submit your manuscript**

QHR is hosted on SAGE Track, a web-based online submission and peer review system powered by ScholarOne Manuscripts™. Visit [http://mc.manuscriptcentral.com/qhr](http://mc.manuscriptcentral.com/qhr) to login and submit your article online. Each component of the manuscript is uploaded separately: Title page, main document, tables, figures, supplemental material.

IMPORTANT: Please check whether you already have an account in the system before trying to create a new one. If you have reviewed or authored for the journal in the past year it is likely that you will have had an account created. For further guidance on submitting your manuscript online please visit ScholarOne.

5.2 **Title, keywords and abstracts**

Please supply a title, short title, an abstract and keywords to accompany your article. The title, keywords and abstract are key to ensuring readers find your article online through online search engines such as Google. Please refer to the information and guidance on [How to Help Readers Find Your Article](https://sagepub.com/authors/how-to-help-readers-find-your-article) in the SAGE Journal Author Gateway on how best to title your article, write your abstract and select your keywords.

5.3 **Corresponding author contact details**

Provide full contact details of the corresponding author including email, mailing address and phone number. Academic affiliations are required for all co-authors. Present these details on the title page, separate from the article main text, to facilitate anonymous peer review.
6. On acceptance and publication

6.1 Fees
There are no fees to submit or publish, unless an author chooses to publish with open access. See “Open Access and SAGE Choice” below. Fees for color reproduction of figures in print may also apply.

6.2 SAGE Production
Your SAGE Production Editor will keep you informed as to your article’s progress throughout the production process. Proofs will be sent by PDF to the corresponding author to make final corrections and should be returned promptly.

6.3 Access to your published article
SAGE provides authors with online access to their final article. There is no set time frame when an article will be assigned to an issue.

6.4 OnlineFirst publication
OnlineFirst allows final revision articles (completed article in queue for assignment to an upcoming issue) to be published online prior to their inclusion in a final print journal issue, which significantly reduces the lead time between submission and publication. Articles published OnlineFirst are assigned a DOI number, but no volume/issue/page number information. Articles will be searchable in PubMed but the citation will not appear with volume/page number information until officially published in an issue. For more information, please visit our OnlineFirst Fact Sheet.

6.5 Open Access and SAGE Choice
Articles accepted in QHR have the option to be published as open access after payment of an article processing charge (APC) paid by either the funder or author. Authors wishing to publish open access should contact openaccess@sagepub.com to make the request. Read SAGE Choice FAQs here.

7. Further information
Any correspondence, queries or additional requests for information on the manuscript submission process should be sent to the QHR editorial office as follows:

Vanessa Shannon, Managing Editor, vshannonqhr@gmail.com
14 August 2014

Mrs Joanne Middleton
Division of Clinical Psychology
2nd Floor, Zochonis Building, Bruswick Street
University of Manchester
M13 9PL

Dear Mrs Middleton

<table>
<thead>
<tr>
<th>Study title:</th>
<th>Illness related communication between parents and children with sickle cell disease: Parental experiences and support needs</th>
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<tbody>
<tr>
<td>REC reference:</td>
<td>14/WM/1093</td>
</tr>
<tr>
<td>IRAS project ID:</td>
<td>147378</td>
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</table>

The Proportionate Review Sub-committee of the NRES Committee West Midlands - Solihull reviewed the above application on 13 August 2014.

We plan to publish your research summary wording for the above study on the NRES website, together with your contact details, unless you expressly withhold permission to do so. Publication will be no earlier than three months from the date of this favourable opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to make a request to postpone publication, please contact the REC Assistant Joanne Unsworth, nrescommittee.westmidlands-solihull@nhs.net.

**Ethical opinion**

On behalf of the Committee, the sub-committee gave a favourable ethical opinion of the above research on the basis described in the application form,
protocol and supporting documentation, subject to the conditions specified below.

**Conditions of the favourable opinion**

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

- Change the name of the NRES Committee in the Participant Information Sheet from Greater Manchester to Solihull.
- Change any mention of tape-recording to audio-recording in the Participant Information Sheet and Consent Form.

You should notify the REC in writing once all conditions have been met (except for site approvals from host organisations) and provide copies of any revised documentation with updated version numbers. The REC will acknowledge receipt and provide a final list of the approved documentation for the study, which can be made available to host organisations to facilitate their permission for the study. Failure to provide the final versions to the REC may cause delay in obtaining permissions.

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

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

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

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

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

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

**Registration of Clinical Trials**

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database within 6 weeks of
recruitment of the first participant (for medical device studies, within the timeline determined by the current registration and publication trees).

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to contest the need for registration they should contact Catherine Blewett (catherineblewett@nhs.net), the HRA does not, however, expect exceptions to be made. Guidance on where to register is provided within IRAS.

**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).**

**Ethical review of research sites**

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

**Summary of discussion at the meeting**

The PRS Sub-Committee confirmed the study raised no material ethical issues under the following headings: Social or scientific value; scientific design and conduct of the study, recruitment arrangements and access to health information, and fair participant selection, favourable risk benefit ratio; anticipated benefit/risks for research participants (present and future), care and protection of research participants; respect for potential and enrolled participants’ welfare and dignity, suitability of the applicant and supporting staff, Independent review, suitability of supporting information, other general comments and suitability of research summary

**Ethical issues raised, noted and resolved in discussion:**

**Informed consent process and the adequacy and completeness of participant information**
The Sub-Committee noted that the name of the NRES Committee reviewing the study was incorrect in the Participant Information Sheet.

The Sub-Committee commented that ‘audio-recording’ would be a better term than ‘tape-recording’ when referring to recording the interviews in the Participant Information Sheet and Consent Form.

**Approved documents**

The documents reviewed and approved were:

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<thead>
<tr>
<th>Document</th>
<th>Version</th>
<th>Date</th>
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<td>Interview schedules or topic guides for participants [Appendix F sickle cell interview schedule]</td>
<td>1</td>
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</tr>
<tr>
<td>IRAS Checklist XML [Checklist_05082014]</td>
<td></td>
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<tr>
<td>Participant information sheet (PIS) [Appendix B Participant information sheet]</td>
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<tr>
<td>Summary CV for supervisor (student research) [Fiona Ulph CV]</td>
<td>1</td>
<td>04 August 2014</td>
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**Membership of the Proportionate Review Sub-Committee**

The members of the Sub-Committee who took part in the review are listed on the attached sheet.

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

**After ethical review**

**Reporting requirements**

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

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

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

**User Feedback**

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website: [http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/](http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/)

**HRA Training**

We are pleased to welcome researchers and R&D staff at our training days – see details at [http://www.hra.nhs.uk/hra-training/](http://www.hra.nhs.uk/hra-training/)

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

<table>
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<tr>
<th>14/WM/1093</th>
<th>Please quote this number on all correspondence</th>
</tr>
</thead>
</table>

Yours sincerely

pp.
Dr Rex J Polson Chair

Email: nrescommittee.westmidlands-solihull@nhs.net

Enclosures: List of names and professions of members who took part in the review Copy to: Ms Lynne Macrae
Dr Lynne Webster, Head of Research Office, Central Manchester University Hospitals NHS Foundation Trust
NRES Committee West Midlands - Solihull

Attendance at PRS Sub-Committee of the REC meeting on 
13 August 2014

Committee Members:

<table>
<thead>
<tr>
<th>Name</th>
<th>Profession</th>
<th>Present</th>
<th>Notes</th>
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<tbody>
<tr>
<td>Dr Richard Mupanemunda</td>
<td>Consultant Paediatrician</td>
<td></td>
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<tr>
<td>Dr Rex J Polson</td>
<td>Consultant Physician - Chair</td>
<td>Yes</td>
<td></td>
</tr>
<tr>
<td>Dr Timothy Priest</td>
<td>Consultant in Pain Management - Vice Chair</td>
<td>Yes</td>
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Also in attendance:

<table>
<thead>
<tr>
<th>Name</th>
<th>Position (or reason for attending)</th>
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<tbody>
<tr>
<td>Miss Andrea Graham</td>
<td>Deputy Regional Manager</td>
</tr>
<tr>
<td>Ms Ellen Swainston</td>
<td>REC Manager</td>
</tr>
<tr>
<td>Miss Joanne Unsworth</td>
<td>REC Assistant</td>
</tr>
</tbody>
</table>
18 August 2014

Mrs Joanne Middleton
Division of Clinical Psychology
2nd Floor, Zochonis Building, Bruswick Street
University of Manchester
M13 9PL

Dear Mrs Middleton

| Study title: | Illness related communication between parents and children with sickle cell disease: Parental experiences and support needs |
| REC reference: | 14/WM/1093 |
| IRAS project ID: | 147378 |

Thank you for your letter of 15 August 2014. I can confirm the REC has received the documents listed below and that these comply with the approval conditions detailed in our letter dated 14 August 2014

Documents received

The documents received were as follows:

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</table>
Approved documents

The final list of approved documentation for the study is therefore as follows:

<table>
<thead>
<tr>
<th>Document Description</th>
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</thead>
<tbody>
<tr>
<td>sickle cell interview schedule</td>
<td></td>
</tr>
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</tbody>
</table>

You should ensure that the sponsor has a copy of the final documentation for the study. It is the sponsor’s responsibility to ensure that the documentation is made available to R&D offices at all participating sites.

14/WM/1093 Please quote this number on all correspondence

Yours sincerely

Ellen Swainston REC Manager

E-mail: nrescommittee.westmidlands-solihull@nhs.net

Copy to: Ms Lynne Macrae

Dr Lynne Webster #
Appendix 5: Local research and development approval letter

Central Manchester University Hospitals NHS Foundation Trust

Research Office
1st Floor, The NOWGEN Centre
29 Grafton Street
Manchester M13 0UU
Tel: 0161-276-3565

Mrs Joanne Middleton
Division of Clinical Psychology
2nd Floor, Zochonis Building, Bruswick Street
University of Manchester
M13 8PL

7th September 2015

Dear Mrs Middleton

PIN: R04084
REC Reference: 14/WM/1093
Research Study: Illness related communication between parents and children with sickle cell disease: Parental experiences and support needs

Thank you for submitting the above study for NHS R&D permission. The University of Manchester is the Sponsor for this study which is not on the NIHR portfolio.

I am pleased to confirm that the Research Office has now received all necessary documentation, and the appropriate governance checks have been undertaken. This letter is issued subject to the research team complying with the attached conditions, Trust SOPs, the DH Research Governance Framework, and any other applicable regulatory requirements. This approval is in relation to the documentation listed.

Please note that Verna Davis cannot take part in research activities until documentation of Good Clinical Practice training is obtained.

I would like to take this opportunity to wish you well with your research.

Yours sincerely

[Signature]

Dr Max Pilotti
Research Contracts Manager

cc. Alison Robinson
Appendix 6: Participant Information Sheet

Study title: Illness related communication between parents and children with sickle cell disease: Parental experiences and support needs

You are being invited to take part in a research study. Before you decide whether you would like to take part it is important for you to understand why the research is being done and what it will involve. Please take time to read the information carefully and discuss it with others if you wish. Please ask if there is anything that is not clear or if you would like more information.

Why is the research taking place?
Parents of children with sickle cell disease are encouraged to be involved in managing their child’s condition at home. This involves knowing about sickle cell disease and feeling able to talk about it with their child. However, little is known about parents’ experiences of this.
This study will involve interviewing parents about their experiences of talking with their child about sickle cell. The findings will add new information about parent’s experiences and will also inform services on how best to support parents.

Why have I been chosen?
You are being invited to take part as you are the parent of a young child with sickle cell disease who attends the paediatric haemoglobinopathy clinic at Royal Manchester Children’s Hospital. We are looking for 25-30 parents to take part in the study.

What will happen to me if I take part?
If you do decide to take part, please keep this information sheet and return the consent to contact form. If you decide to take part, the researcher will contact you in order to arrange an interview. The interviews will take place at your home or at the Sickle Cell and Thalassaemia centre in Manchester.

The interview will last between 1 and 1½ hours. All of the questions will be about how you and your child talk about sickle cell disease. The interview will be audio recorded, but you will be free to stop the recording at any time. Audio recording the interview is important so that the researchers have an accurate recording of what you say.
What if I decide I don’t want to take part?
It is your choice whether you take part or not and nobody will be upset if you decide not to take part. Choosing not to take part will not affect the standard of care you, or your child, receives, now or in the future. If you wish to withdraw from the study at any point just tell the researcher that you do not wish to continue. We will destroy identifiable information and the data collected up to your withdrawal.

What are the possible disadvantages and risks of taking part?
Researchers are always required to tell you about any risks to you, should you agree to take part in research. It is possible that talking about your child’s condition may upset you, and if at any point you felt uncomfortable or upset during the interview, you would be free to stop the interview.

What are the possible benefits of taking part?
Although this research won’t benefit your child directly, you may find it helpful to share your experiences with the researcher and have your experiences heard. We hope that the findings of this study will allow health care professionals to support parents in coping with the challenge of talking to their child about sickle cell disease.

Will my taking part in this study be kept confidential?
Your information and the recordings from the interview will be kept strictly confidential and will only be used for the purpose of this study. The recordings from the interview will be kept on a computer and protected by passwords that are known only to the researcher. The recordings will be transcribed (typed out) by the researcher and will not include any information that would identify you. After this the recording will be deleted from the computer. The transcription of the interview will be kept securely on a password protected document on a secure University network drive until a maximum of 5 years after the study. It will then be destroyed. Study data and material may be looked at by individuals from the University of Manchester, from regulatory authorities or from the NHS trust, for monitoring and auditing purposes, and this may well include access to personal information.

What will happen to the findings of the research study?
The findings will be published in academic articles, but there will be no identifying information from any of the participants in the articles. Any quotes that are used in the article will be quoted under a false name. When the study is finished, the parents who have taken part in the research will be sent a short summary of the results.

Who is organising the research?
The research is being organised by the University of Manchester, in collaboration with Central Manchester University Hospitals NHS Foundation Trust. Joanne Middleton is conducting the study as part of a Clinical Psychology Doctorate (ClinPsyD) qualification at the University of Manchester and is working under the supervision of Dr Fiona Ulph and Professor Rachel Calam. Dr Sonia Patel from the Sickle Cell and Thalassaemia Service is also involved in this research.
Who has reviewed the study?

This study has been reviewed by NRES Committee West Midlands - Solihull and was given a favourable opinion on 13.8.2015. The REC reference is 14/WM/10.

What if there is a problem?

In the event that something does go wrong and you are harmed during the research you may have grounds for a legal action for compensation against the University of Manchester or Central Manchester University Hospitals Foundation Trust but you may have to pay your legal costs. The normal National Health Service complaints mechanisms will still be available to you.

If you have a concern about any aspect of this study, you should ask to speak to the researchers who will do their best to answer your questions. If they are unable to resolve your concern or you wish to make a complaint regarding the study, please contact a University Research Practice and Governance Coordinator on 0161 2757583 or 0161 2758093 or by email to research.complaints@manchester.ac.uk

What do have to do if I want to take part?

If you have decided that you would like to take part, please return the Consent to Contact form and the researcher will be in touch to arrange an interview and answer any questions you might have.

Thank you for considering taking part in this study.

Joanne Middleton
Trainee Clinical Psychologist
The University of Manchester
Division of Clinical Psychology
Manchester M13 9PL
joanne.middleton@postgrad.manchester.ac.uk

Professor Rachel Calam
Professor of Child and Family Psychology
School of Psychological Sciences
Manchester M13 9PL
Telephone: 0161 306 0403
rachel.calam@manchester.ac.uk

Dr. Fiona Ulph
Lecturer in Qualitative Methods
School of Psychological Sciences
University of Manchester
Manchester M13 9PL
Telephone: +44 (0)161 275 1979
fiona.ulph@manchester.ac.uk

Dr. Sonia Patel
Paediatric Psychosocial Department
Royal Manchester Children's Hospital
Telephone: 0161 701 0850
sonia.patel@cmht.nhs.uk
Appendix 7: Consent to Contact form

Consent to contact form

Study Title: Illness related communication between parents and child with sickle cell disease: Parental experiences and support needs

I am happy for a researcher to contact me to discuss taking part in the above study.
Name..........................................................................................................................
Signature..................................................................................................................

My contact details are:
Telephone number..................................................................................................

The best time to contact me is (tick all that apply):
Between 9-5 ☐
After 5 pm ☐
Weekdays ☐
Weekend ☐

Please return this form to the researcher in clinic or in the stamp addressed envelope provided.
Appendix 8: Consent Form

Study Title:
Illness related communication between parents and children with sickle cell disease: Parental experiences and support needs

Please Initial Box

1. I have read and understood the information sheet dated: 05/08/2014 (version 2) for the above study. I have had the chance to ask questions and have had these answered clearly.

2. I understand that my participation is voluntary and that I am free to withdraw at any time, without having to say why.

3. I give my permission for a typed version of the interview to be Kept by the researcher and used anonymously in connection with the study.

4. I give my permission for the interview to be audio recorded and for the use of anonymous quotes to be used in publications.

5. I understand that information collected during the study may be looked at by individuals from the University of Manchester, from regulatory authorities or from the NHS Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my data.

6. I would like to receive a summary of the findings from the study.

7. I agree to take part in the above study.

Name of participant……………..Signature:………………Date:……

Name of person taking consent…Signature:………………Date:……
Appendix 9: Interview Schedule

**Interview schedule**

**Opening/Introduction to the interview**
In this research we’re talking to parents of young children with sickle cell disease. We’re interested in the conversations you have with your child about their condition. Today I’ll be asking about how you and your child talk about them having sickle cell and about whether there are things that are difficult that you’d like more help with. There are no right or wrong answers or ways of saying things. What’s important is that we hear your experiences of talking about sickle cell with your child.

**First conversations about sickle cell**
Take me back to when you first talked to your child about them having sickle cell. Tell me what happened.
- What prompted the conversation?
- Can you tell me about any planning that was done about the conversation?
- Before it happened, how did you feel about having that conversation?
- How did you tell them?
- How did your child respond?
- Did they ask questions?
- What questions?
- How did you answer them?
- What was it like for you having this conversation?
- How did you feel after the conversation?
- Would you have changed anything about it? What would you have changed?

**Later conversations**
What conversations can you remember soon after you first talked about it?
Are there any that stick out in your mind?
What were they about?
If they happen often:
- Do the conversations tend to be similar are they different each time?
- If similar- what do they tend to be about? Who tends to start them?

**Child’s understanding of sickle cell / development of their understanding over time**
What do you think your child understands about sickle cell disease?
Has this changed over time? What change have you noticed?
What do you think has led to this change?

**Questions for parents with sickle cell:**
You’ve told me that you have sickle cell disease yourself. Do you talk about your own condition with your child?
If yes:
What have you talked about?
What is it like to talk with your child about living with sickle cell?
If no:
What are the reasons for not talking about it?

Illness specific questions:
I’m aware that children with sickle cell can have lots of hospital appointments. Could you tell me about the conversations you have with them about their hospital appointments?
  For example, do you talk about the appointment beforehand? What do you talk about?
  What about after the appointment? What do you talk about then?
Coping questions: I’d like to hear about if and how you talk to your child about their symptoms:
  What kinds of symptoms of sickle cell does your child get?
  How do you think they feel when they have these symptoms?
  What do you say to them when they have these symptoms?
  What happened the last time your child was ill?
  What did you talk about together?
  What about talking with them about any negative feelings they have about sickle cell? Does this happen?
  If so:
  What is that like? What do you say to them when they say negative things about having sickle cell?

Support needs
Are there things about sickle cell that you feel are difficult to talk about with your child? What are they?
What would make these conversations easier?
Is there anything that services could do or provide to help?
  (e.g. prompt- Leaflets, Support groups, parent education groups, individual consultations with specialist nurse/ psychologist)

We’re coming to the end of the interview now....
Is there anything that I haven’t asked about that you would like to add?

If you have one message about what it’s like to talk to children about sickle cell what would it be?

Do you have any questions for me?

Thank you for your time and your interest in the research. If the interview has raised any issues that you would like to talk about, I have attached the contact details of the parents’ support group at the sickle cell service and a list of other organisations that might be able to help. If you have any other questions about the research itself, please do not hesitate to contact me on the details given on the information sheet.
Appendix 10: Condensed version of the interview schedule

Interview Prompt sheet

Setting things up
Explain research. No right or wrong answers. Hear your experiences.
Strange conversation. Lots of me nodding and listening.
Notes. Repeat, louder, clearer if needed for tape.

First conversations
Take me back to when you first talked.
Any planning?
How did it go? How child respond? Questions? Your answers?
How did you feel? Before, after, during
Changed anything?

If can’t remember:
When have you talked about?
Tell me about when you talk?
When was the first time? How old?

Later conversations
Conversations soon after? Any stick out?
How often do you talk now?
Often: what about? How start?

Not Talking
What do you talk about/ What don’t you talk about?
What is not said/ talked about?
What are your reasons for not talking about SCD?
What do you do/ say instead?

Child’s understanding – what do you think they understand? How changed?
If SC: Do you talk about your condition? Yes What have you talked about? What is it like?
No: Reasons for not?

Appointments:
Conversations about their hospital appointments? Talk before/after? about?

Talking about symptoms:
What kinds of symptoms? What do you say to them?
Last time your child was ill? What did you talk about together?
Any negative feelings? What is that like to talk about? What do you say?
Support
What’s difficult to talk about? What would make it easier? Anything services could do/provide?

Closing
Coming to the end now…Anything to add? Questions for me?
One message, what would it be?

What will happen next?
Thank you letter- contact details (re: research)
Type up interview. Analyse and write up.
Another letter to you with a summary
Participant Questionnaire

Study title: Illness related communication between parents and children with sickle cell disease: Parental experiences and support needs

This questionnaire collects information about you and your family. Please read and answer each question.

1. Your age: ______ (years)

2. Child's gender: Male □ Female □

3. Child's age: ______ (years)

4. Which of the following sickle cell disorders does your child have?
   - Sickle Cell Anaemia (HbSS) Yes □ No □
   - Sickle Cell Haemoglobin C Disease Yes □ No □
   - Hemoglobin S-beta-thalassemia Yes □ No □
   - Other form of sickle cell disease Yes □ No □

5. Your relationship to this child:
   - Mother (biological or adoptive) □
   - Step-mother □
   - Foster mother □
   - Father (biological or adoptive) □
   - Step-father □
   - Foster father □
   - Other (please describe) ________________________________

6. Your current marital status:
   - Married □
   - Divorced/separated □
   - Single □
   - Cohabitating □
   - Widow/er □
   - Other (please describe) ______

7. Which ethnic or cultural group do you most strongly identify with? (please write your response in the space) __________________________
8. Your highest level of education:

- primary school or less
- some high school
- completed high school
- trade/technical college qualification
- university degree
- post-graduate degree

9. Are you working outside the home right now?

- yes, full time
- yes, part time
- not working, but looking for a job
- home based paid work (child care, internet or phone-based work, etc)
- not working (includes stay at home parents, retired)

10. Is your partner working outside the home right now? (if applicable)

- yes, full time
- yes, part time
- not working, but looking for a job
- home based paid work (child care, internet or phone-based work, etc)
- not working (includes stay at home parents, retired)