

Children's Perspectives on Living with a Sibling with a Chronic Illness

Antoinette Deavin BSc, MRes, DClinPsy, Pete Greasley BSc, PhD, Clare Dixon BSc, DClinPsy, PhD

Affiliation: Lancaster University, Lancaster, United Kingdom

Address correspondence to: Pete Greasley, Department of Clinical Psychology, Furness Building, Lancaster University, Lancaster, LA1 4YG, 01524 593535, p.greasley@lancaster.ac.uk.

Short Title: Living with a sibling with a chronic illness.

Funding Source: No funding was secured for this study.

Financial Disclosure: The authors have no financial relationships relevant to this article to disclose.

Conflict of Interest: The authors have no conflicts of interest relevant to this article to disclose.

Abbreviations: Paediatric Chronic Illness (PCI)

Table of Contents Summary

This review synthesises the experiences of healthy siblings living with a sibling who has a paediatric chronic illness, producing two overarching themes and five sub-themes.

Contributors' Statement:

Dr Antoinette Deavin conceptualised and designed the study, carried out the synthesis and drafted the initial manuscript.

Drs Greasley and Dixon reviewed the initial concept, data collection and analysis, and critically reviewed and edited the manuscript.

All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

Abstract

Context: Clinical guidance emphasises the importance of considering the whole family when caring for a child with a paediatric chronic illness (PCI). However, there is a lack of research specific to sibling experience. Studies utilise parental accounts to examine family experiences and consequently the direct voices of siblings tend to be neglected.

Objective: The meta-synthesis explored the experiences of siblings of children with PCIs to understand their perspectives; in particular what they feel had influenced their emotional wellbeing, in order to target interventions.

Data Sources: PsychInfo, CINAHL, PubMed, and Academic Search Complete databases searched from inception to April 2016.

Study Selection: Qualitative articles examining children's perspectives of living with a sibling with a PCI. 12 articles reviewed.

Data Extraction: Participants characteristics, qualitative methodology, analysis and themes.

Results: The thematic synthesis identified two overarching themes providing new insights. The first theme was 'Changing relationships' which had two sub-themes: 'Changing family relationships' and 'Changing relationship to self'. The second theme was 'Managing changes' which had three sub-themes: 'Coping and acceptance', 'Support from friends, peers and support groups' and 'Negative reactions from others'.

Limitations: Studies were often descriptive with little explicit qualitative analysis.

Conclusions: The findings outline how changes in family relationships often result in reduced communication and a suppression of healthy siblings' needs. Siblings develop strategies to help them cope with and accept their circumstances, including finding new prosocial ways of meeting their needs in the form of skills and roles they develop.

There is an emotional toll that families experience from living with a child who has a PCI and may frequently be unwell or in pain, or struggling to adjust to their condition.^{1,2} These experiences impact on the quality of life of the individuals who have the illness and family members.³ This is recognised in professional guidance for recommendations of family based care and interventions, where the needs of each family member are considered and supported.^{4,5} However, within these guidelines the discussion of siblings is marginal. This is also true of the representation of siblings in research, which generally focuses on overall family experiences or prioritises parental accounts of sibling experiences.⁶⁻¹⁶ Consequently the voices of siblings are overlooked. It has been commented that the effect on siblings has received little attention¹ and it is difficult to make clinical recommendations based on available research.¹⁷

Studies suggest increased levels of sibling distress compared to peers,¹² however results have often been mixed with contradictory findings. In their review from 2002 Sharpe and Rossiter¹¹ reiterate Cuskelly's statement of how previous research into psychological adjustment of healthy siblings leaves the reader with "...the overwhelming impression of contradiction and confusion."¹⁸ (p.111) Contradictions may reflect the difficulty in consolidating data for different samples (e.g. age, paediatric condition), differing data sources (sibling, parent, teacher), and the wide range of psychosocial constructs used to determine the current psychological state of the well sibling.

Qualitative research has been utilised to understand siblings' experiences, but this has mostly been from a whole family perspective, where all family members are interviewed together. This approach has been used to explore treatment demands,¹⁹ family dynamics,²⁰ perceptions

towards genetic testing²¹ and general psychosocial impacts.¹⁶ While this may help to provide an understanding of family perspectives, sibling's views may be influenced by other members of the family, therefore failing to identify the distinct voices of the siblings. Many studies include siblings of younger ages¹⁶ and, whilst having an adult present and/or their perspective included in the research ensures initial insights into a difficult to engage sample, this may mean that siblings provide socially desirable responses.²⁰ The benefit of this approach is that siblings at least have a partial voice which has provided insight into reactions on diagnosis, education on the chronic illness, sibling's involvement in care, sibling relationships, fears and the overall impact of the disease on the sibling.¹⁶

To further advocate siblings' experiences this synthesis focuses on siblings' direct reports. For some specific diseases, for example paediatric cancer, the sibling experience is investigated with siblings directly and is generally better documented and understood as demonstrated by three reviews.²²⁻²⁴ However it has been argued that a non-diagnosis based review could focus on the psychosocial commonalities that would help to improve care for families.¹ A more generic review with the intention of unifying siblings' voices across diseases has not yet been undertaken. Thus, the aim of this meta-synthesis of qualitative research was to fill this gap and address the research question: What are the experiences of siblings of children with a chronic and non-communicable physical health condition?

Method

Database Searches

Four databases were searched: PsychInfo, CINAHL, PubMed, and Academic Search Complete. Search terms were generated from APA recommended vocabulary, each

database's specified term generator, key words from relevant articles, and reviewing search terms with the research team (Table 1).

[Table 1]

Inclusion Criteria

- The PCI that affected the unwell sibling must have been physical, chronic, non-communicable, incurable, and require ongoing care.
- Studies reporting a qualitative approach which elicited first-hand accounts, in the form of interviews or written responses e.g. open-ended questionnaires, and where the sibling's experiences could be identified independently in the findings i.e. as a separate theme.
- The content of the interview had to discuss the siblings' experiences as children (under the age of 20 years old to account for all childhood experiences)
- Published in English before April 2016.

Exclusions:

- Articles that targeted disease-specific experiences or procedure (e.g. being a donor for a hematopoietic stem cell transplant)²⁵ as part of the research question were excluded.
- Experiences relating to physical injury (e.g. brain injury), mental health or addiction were not included.
- Papers relating to cancer. Some forms of cancer are recognised as being acute e.g. acute leukaemia, and treatment in other forms can lead to long-term remission of the symptoms, which was felt to make this disease sufficiently different from the sample to exclude childhood cancer from this review.

Selection Process

Articles were identified, screened and selected using the inclusion/exclusion criteria as outlined in Figure 1. This produced the final sample of 12 articles. Summaries of the aims, key features and themes of the final sample are provided in Table 2.

[Figure 1]

[Table 2]

Quality/Critical appraisal

The Critical Appraisal Skills Programme²⁶ qualitative checklist was used for quality appraisal of the studies (Table 3). CASP provides 10 questions relevant to appraising qualitative studies (e.g. Was the data analysis sufficiently rigorous?), to which a rating of 0 (no evidence present), 1 (some evidence), 2 (good evidence), 3 (very good evidence) was applied, with a maximum score of 30 for each paper. Scores were assessed independently by the first author and a colleague, both experienced in qualitative research, and then averaged to provide a guide to the quality of the reported studies. From Table 3 we can see that there were no significantly weak studies, with ratings ranging from 13-23.5 (mean 18.9).

[Table 3]

Meta-synthesis

The thematic synthesis²⁷ followed three main stages: 1) identifying codes directly from the data in the studies; 2) using these codes to generate initial categories and themes; 3) using these codes, categories and themes to generate the analytical themes and sub-themes represented in the overarching model of the synthesis (Figure 2). The benefits of this approach are that the analysis remains faithful to the original articles, whilst allowing a transparent deconstruction (coding), synthesis of the data (descriptive themes) and translation

of the concepts (analytical themes).²⁷ The discussion sections were also reviewed to check for new information, however codes were only included when it was grounded in the data rather than conceptual interpretations.

[Figure 2]

Findings

From Figure 2 we can see that the systematic review yielded two major themes: '*Changing relationships*' and '*Managing changes*'. '*Changing family relationships*' occurred between all members of the family as a result of their changing circumstances and their own emotional responses. This led to a 'changing relationship to self' for siblings as a result of meeting the changing needs of the family and consequently acquiring new roles, skills and responsibilities which frequently became part of their long-term identity. Managing these practical and psychological changes (theme 2) requires siblings to develop ways of coping, accepting and adjusting, which is often influenced by external support or their fear of negative reactions from others.

Theme One: Changing Relationships

Subtheme One: Changing Family Relationships

This subtheme outlines siblings' perceptions of changes to family cohesion, changes to their relationships with their parents and with their unwell sibling.

An altered sense of family cohesion since the onset and diagnosis of the disease was reported in five articles. Siblings expressed views representing both a sense of increased and decreased family cohesion. When positive experiences were reported, siblings described a united family

environment,^{28,29} with the whole family contributing to their unwell sibling's care,^{29,30} which they felt brought them closer²⁹ giving the family something in common³¹ which they shared and talked about together.³⁰ Participants felt this level of closeness and bonding was different to other families that did not have to manage a PCI.^{28,31} Findings from ten papers addressed the changing relationship between parents and their healthy children. These highlight changes in the amount and quality of parental attention given to each of the children,^{29,31-37} the perceived change in parental expectations of the healthy sibling (e.g. helping to care for the unwell sibling),^{28,35} and a reduced level of communication with their parents.^{29,35,36} The majority of reports from healthy children describe a shift of parental attention to their sibling. They often felt jealous and resentful of the amount of time, protection and 'special treatment' parents bestowed upon their siblings,^{31,32,37} especially when their sibling appeared to be 'doing well',³⁵ although they acknowledged the increased attention was necessary.³²

Healthy siblings from two studies described what has been termed as 'parental silence', whereby parents do not talk to them about the illness in order to protect them from the difficult aspects of the disease.^{35,38} This meant parents sometimes did not know how much siblings knew about the illness.³⁸ Participants in the study by Wennick and Huus²⁹ discussed how siblings would return the silence as their parents had 'enough to worry about'. This resulted in a reciprocal silence between parents and the healthy sibling,³⁵ which is potentially problematic for family relationships as some participants reflected that openness might provide the antidote to family breakdown.³⁷

The relationship between siblings appeared to be equally complex, eliciting a range of emotional responses. For example, the increased time spent together meant some had a closer relationship,³⁰ but six studies found that healthy siblings recognised negative changes in their

sibling relationship, describing it as a loss³¹ or distancing.³⁰ These were associated with changes in their unwell sibling,^{32,37} for example becoming more prone to mood swings.²⁹

Families hold varying beliefs about how to view and approach illness as a unit and as individuals.¹⁴ For some the illness is all but ignored in attempts to create a sense of self that is unaffected, but for others they incorporate the illness in the new family identity to the point where it may become the sole focus, causing the family to define themselves based on providing care.³⁹ The sense of increased family cohesion some children mentioned may therefore result from identifying with the family system's roles and agendas, causing them to feel included. However, this presumptive allocation of roles has repercussions for siblings who do not fit with the 'acceptable' assumptions of the family system. For example, one common and potentially damaging issue is the reciprocal silence that occurs when familial beliefs promote a lack of discussion about the illness, in order to protect others or because it is too painful. This causes siblings who want to discuss their emotions to feel as if they are threatening the family, perhaps becoming a burden, forcing them to become isolated and unable to address their needs. These internal system boundaries,³⁹ set up to protect each other, consequently reduce communication and cause members to become distant at times when they need each other the most. Given that the structure and hierarchy of the system is also governed by parental beliefs, it is important to remember that not all members have equal power and voice. With healthy siblings being both children and perceived of as being in less urgent need, their voices may often be ignored and suppressed by the adults around them.

Subtheme Two: Changing Relationship to Self

As a result of the changes in the family, healthy siblings became more aware of their role within the family and felt forced to change. All the articles acknowledged the changes

occurring within the family had an impact on the emotional experiences of healthy siblings. Two studies^{28,32} noted there were positive impacts including a positive sense of self and their life experiences, but the majority discussed the overwhelming sense of negativity. In Derouin and Jessee's article³² some siblings went so far as to cite themselves as the "most unhappy member of the family"^(p142). Their empathy with their sibling often led to sadness that they had the disease or anger when others bullied their sibling because of the disabilities arising from the PCI.^{28,35,37}

Worrying about their sibling's health and wellbeing was pervasive.^{31,32,35,37,38} Some recognised that worrying had begun to interfere with other areas of their life, particularly causing problems with concentration at school.³⁸ Siblings also worried about the uncertainty of their future.³⁷ Dependent on the type of illness their sibling was diagnosed with, participants reported anxiety about the development and transmission of the illness, specifically how it might affect their health^{31,34} and whether they would be a carrier for their children.^{30,35} Other emotions reported in the literature were feelings of "survivor guilt",^{28,34,37} loneliness resulting from familial and peer isolation^{28,35,37,38} and jealousy due to the reduced level of parental attention.^{30,34}

Findings from Read et al³⁰ described the difficulties participants experienced in balancing home demands, including parental and sibling expectations, whilst their own needs were often overlooked. Given siblings' perceptions that their needs were not as important as their ill siblings, it is not surprising participants in five articles stated they did not discuss their feelings with their family for fear of their parents becoming angry, increasing their parents' worry³⁴ and adding to their burden.³⁸ In addition, siblings discussed avoiding conflict^{35,40} and pleasing family members to gain approval.³⁵ The desire to balance the needs of the

family, leading to avoidance of communicating their emotions, appears to provide the foundation of siblings' experiences of not recognising,³⁵ deprioritising^{34,40} and suppressing their own needs, with a desire to be emotionally self-sufficient.⁴⁰ Bradford's² hypothesis of "sibling self-sustainability" due to reduced parental attention is synonymous with this sense of emotional self-sufficiency. "Self-sufficiency" is thought to be promoted by reduced communication from parents in order to protect the sibling. Furthermore, siblings regularly do not have direct contact with a health professional, compounding the lack of communication and need to be self-sustaining.⁴¹

In all of the articles healthy children described the new roles and skills they had developed since the onset of their sibling's condition. Participants described how this felt like a role transfer and found themselves embodying roles associated with older adolescents or adults.^{30,37} Their new experiences provided them with specialist knowledge of the condition and the responsibilities placed upon them caused them to develop a caring role, which became a part of their identity.^{28,29,37} These skills and tasks ranged from general caring for the family e.g. housekeeping, supervision of their sibling,^{29,31} helping with leisure activities,³⁰ to identifying, assessing and managing their sibling's symptoms.^{30,33,38} The findings of Brennan et al highlight the extent to which the caring persona was internalised and supported by others, demonstrated by friends calling the healthy sibling "Mum".^{40 (p.819)} One sibling noted how their experience had made them more compassionate towards other people generally.³⁶ Siblings described their caring role with pride, noting the acceptance that came with the role, but also recognising the limitations.^{37,38} The caring role was discussed as increasing over time,³⁰ with few breaks,³⁵ and some discussed the shame they experienced when they did not meet their own expectations of the role.³⁴

The acquisition of specialist knowledge, skills and internalised positive roles appears to have provided siblings with an identity that makes them feel unique and special;³¹ some acknowledged their experiences shaped their career choices e.g. becoming a family support worker or nurse.³⁰

Theme Two: Managing Changes

This theme outlines how healthy siblings cope with and accept the changes that result from their sibling's disease. It consists of three sub-themes: '*Coping, acceptance and adjustment*', '*Support from friends, peers and support groups*' and '*Negative reactions from others*'.

Subtheme One: Coping, Acceptance and Adjustment

Adjustment over time was a factor described in five articles as having an influence on coping and acceptance. Siblings described the impact of the illness as getting easier over time.³¹ They commented that it was harder when their sibling was first diagnosed²⁸ but, where there was a slow progress of the disease this allowed coping and acceptance.³⁰ Time was felt to create space to make sense,²⁸ allowing the process of accommodating to their sibling's decline.³⁸

Participants described the evolving process of accruing of information³⁰ and the importance of providing siblings with information about the disease.^{30,35} Some commented that a lack of information, particularly relating to symptom management, led to fear.²⁹ Parents were noted as the main source of information;³⁰ however when they were unwilling to discuss the disease siblings would find alternatives, for example support groups or the internet.^{30,35}

When siblings had access to information they started changing their attitudes towards the disease and its impacts.³⁷ Siblings described how they developed insight and understanding of the broader situation³² and empathy for their family members experience.^{30,31,33,38}

Understanding sometimes helped them to cope with the imbalance of attention between themselves and their sibling.²⁹ For others, they still felt jealousy or neglect but could tolerate and accept the necessity of the situation.³⁰

These findings suggest that siblings would prefer to have increased levels of information earlier relating to the disease and its impacts, which may help build their understanding and empathy, helping them to tolerate and accept the situation and their feelings. This is supported by findings showing strong associations between increased sibling illness knowledge, positive sibling attitude toward the illness and sibling behaviour.⁴² However it is important to note that this depends on the age of the sibling, with older children having more illness knowledge.⁴²

Many articles presented findings relating to behavioural and cognitive strategies that allowed healthy children to cope with and/or accept having a sibling with a PCI. These appear to be either distancing or integrating techniques. Distancing techniques included avoidance,³⁰ distraction,³² physical and temporal compartmentalisation.^{35,40} These were often described as “getting on with it”, “focussing on the day-to-day”, “living in the present” and removing themselves from difficult situations though pursuing external interests³⁷ in order to keep their home and ‘other’ life separate and maintain some level of “normality”. Bellin et al²⁸ noted how siblings deemed “normalising” as a step toward acceptance and integration of the disease into their lives.

Little is documented on the specific coping strategies used by siblings; however, the techniques identified in this meta-synthesis are comparable to those used by children with PCIs. A narrative review by Compas, Jaser, Dunn and Rodriguez⁴³ categorised strategies into three types: active, accommodative or passive coping. Active coping is an individual's attempts to directly influence or change the source of stress. Accommodative coping is an attempt to adapt to the source of stress through "...reappraisal, positive thinking, acceptance or distraction".^{43 (p6)} Passive coping includes cognitive and physical avoidance of the stressor. The review found accommodative coping to be the most effective form of coping, with mixed findings for active coping, and poorer outcomes for those who utilise passive coping techniques.

Subtheme Two: Support from Friends, Peers and Support Groups

Support came from family, friends, teachers and support groups.^{30,38} Findings from Gallo et al³³ detail the internal conflict healthy siblings have when telling other people about their sibling's illness and seeking support - some felt comfortable revealing the illness to others (though some would be selective) but some did not want people to know. Friendships were discussed in five of the articles^{28,30,33,35,38} and were often supportive, mature and inclusive,²⁸ providing both practical and emotional support, though participants from the study by Hutson et al,³⁵ feared that their friends would reject or ignore them due to their siblings. Support groups were mentioned as a form of support that helped them to overcome their isolation and aid their adaptation,³⁰ allowing them to share their experiences and knowledge with each other.³² Such groups enabled connections to others with shared experiences, expanding their social network and offering them more opportunities to make sympathetic friends.³¹

Subtheme Three: Negative Reactions from Others

Findings from seven studies identified the reactions of others to their chronically ill brother or sister as a worry.³⁸ Siblings were worried their ill brother/sister might be teased, rejected by others and become upset,^{28,30,33} even so far as for them to be concerned about prejudice from the community.³⁸ They described becoming embarrassed by the treatment of their brother/sister in public³¹ identifying ignorance as a rationale for their negative behaviour;³⁸ they wished others would be more accepting and tolerant.³⁸

Discussion

The synthesis has found that siblings alter their behaviour in order to meet their own needs and those of the family, taking on tasks and skills that include them in the current caring goals of the family. With time and positive feedback, the behaviours that are reinforced become roles within the family and an internalised positive characteristic of their identity. These characteristics are synonymous with prosocial characteristics listed in the clinical and research literature.^{2,39,40,44,45}

However, this combination of emotional self-sufficiency and increase in prosocial behaviours may lead parents and professionals to perceive the sibling as functioning and thriving. Parents underestimate the emotional responses and needs of healthy siblings,² which may explain why they are frequently overlooked, and why there is a lack of professional guidance regarding the treatment of siblings through the care process.^{41,46} For example, the policy statement from the American Association of Pediatrics highlighted the importance of patient and family centred care, but did not mention specific risks or recommendations for siblings.⁴ Whilst siblings adapt their behaviour to become more prosocial, they are still experiencing high levels of distress; a meta-analysis by Vermaes et al¹² found that siblings had

significantly higher levels of internalising problems (e.g. depression and anxiety) than comparisons.

Siblings have many strategies and resources they use to manage the changes that result from their sibling's disease. Paramount was the access to relevant and timely information, which led to greater understanding and empathy towards family members, and helped them to manage and tolerate their own emotions, in particular their sense of jealousy regarding the imbalance of parental attention. They used a variety of cognitive and behavioural techniques identified in the paediatric patient literature. This review's findings suggest that whilst siblings use active and accommodative coping strategies, many also rely more on passive coping e.g. distancing, which has been linked to poorer outcomes. Sharing their experiences with others could be helpful but there was also the fear of being rejected due to their sibling's illness. Support groups were seen as helpful, but infrequent. Supportive relationships have been helpful for siblings of children with cancer, allowing them to access information to help them make sense of the situation, provide opportunities to communicate how they feel, seek reassurance, receive attention to feel valued and maintain self-esteem and a sense of self independent of the illness.⁴⁷

These findings are comparable to those of children with a sibling with paediatric cancer.^{48, 49.}

⁵⁰ Mixed methodology reviews found that whilst siblings as a whole did not report significantly higher levels of psychiatric disorders, a subset experienced strong negative emotional reactions and their experiences impacted upon their emotional and social wellbeing. More specifically, it was reported that siblings experienced greater distress closer to the time of diagnosis and there were often school difficulties within two years of the diagnosis. Younger siblings often reported somatic complaints, whilst concerns for teenagers

were psychosocial. The qualitative elements of these reviews similarly highlighted the importance of changing family dynamics and routines, siblings' experiences of intense feelings, the development of some positive self-attributes, and recognised their unmet needs. However, this review helps to explain the complex interaction between family dynamics, emotional experiences and changes to the siblings' identity in order to meet their needs. These previously obscured links identify the family's ability, and need, to reduce communication, which often leads to siblings not expressing their needs and conversely appearing to be doing well, which subsequently may explain some of the confusion in research outcomes.

Current good practice guidelines identify the need to consider the entire family when working with families with a PCI.⁴ However there is little guidance as to how this should be implemented with siblings.^{4,5} This synthesis points to several areas for family centred clinical intervention. The findings highlight siblings' desire and ability to conceal their needs and emotions, so professionals should be mindful of their needs and monitor the levels of communication or avenues for support available to the sibling. This could be explicitly addressed by inviting siblings to a hospital appointment where they would have access to a health professional. The professional could answer their questions, discuss age appropriate inclusion of the sibling in caring duties, promote family discussion about the disease and directly discuss the subjective needs of the sibling, which may reduce anxiety regarding treatments and their uncertainties about the future.

Age appropriate leaflets could also normalise the experience for the family. Where possible, families should be encouraged to speak together about the illness with siblings and allow them a forum to express their concerns and help them understand and to feel heard and

included. Keeping channels of communication open would also allow parents to problem solve when necessary. Support groups and other positive events, for example charity work or sibling days⁴⁴ were also found to be helpful but were often too far away for siblings to access regularly. It may be that more online resources and ways of communicating could be developed and encouraged, e.g. sibling forums, buddy systems or websites such as www.sibs.org.uk.

Whilst siblings are proficient at creating a positive prosocial identity, their ability to identify other positive self-attributes are lower than siblings of healthy children.¹² Consequently families and professionals should liaise to ensure that siblings have space and support to develop a positive identity outside of the caring role. At home this could be implemented by ensuring that children are encouraged to have interests and their independent achievements are recognised. More broadly this could be supported by actions to reduce negative reactions within the general population, due to a lack of understanding, such as providing education days at school about chronic illnesses.

Conclusion

This review gives voice to healthy siblings' concerns regarding their inability to disclose their emotions and the necessity to develop skills and roles to meet their own needs by constructing a positive identity that is concordant with the family's needs. Expressing their feelings and needs is hampered by the widening gaps in communication with their support systems, perhaps leading to the clinical symptoms often reported as maladjustment i.e. mood changes, rebellion, attention seeking and somatic complaints.^{14,16,19,51,52} Whilst there is some evidence of considering the clinical importance of siblings' experiences,^{2,39,44} it has been recognised that they have been overlooked within the research literature.^{1,17} This may be as a

result of siblings appearing to be functioning well and in a desired manner to parents and professionals, combined with their fear of expressing how they feel.

Word count: 4326

REFERENCES

1. Wallander JL, Varni JW. Effects of pediatric chronic physical disorders on child and family adjustment. *J Child Psychol Psychiatry*. 1998;39(1):29-46. Available at: <http://www.ncbi.nlm.nih.gov/pubmed/9534085>.
2. Bradford R. *Children, Families and Chronic Disease: Psychological Models and Methods of Care*. London: Routledge; 1997.
3. Alwan A. Global status report on noncommunicable diseases. WHO. 2010. Available at: http://www.who.int/nmh/publications/ncd_report_full_en.pdf. Accessed November 2, 2016.
4. Eichner J, Betts J, Chitkara M, Jewell J, Lye P, Mirkinson L. Patient and family-centered Care and the pediatrician's role. *Pediatrics*. 2012;129:394-404. doi:10.1542/peds.2011-3084.
5. Cancer services for children and young people. NICE. 2014. Available at: <https://www.nice.org.uk/guidance/qs55/resources/cancer-services-for-children-and->
6. Barlow JH, Ellard DR. The psychosocial well-being of children with chronic disease, their parents and siblings: an overview of the research evidence base. *Child Care Health Dev*. 2006;32(1):19-31. doi:10.1111/j.1365-2214.2006.00591.x.
7. Jackson C, Richer J, Edge J. Sibling psychological adjustment to type 1 diabetes mellitus. *Pediatr Diabetes*. 2008;9(4):308-11. doi:10.1111/j.1399-5448.2008.00385.x.
8. Lavigne J, Faier-Routman J. Psychological adjustment to pediatric physical disorders: a meta-analytic review. *J Pediatr Psychol*. 1992;17(2):133-57. doi:10.1093/jpepsy/17.2.133.
9. LeBovidge JS, Lavigne J, Donenberg GR, Miller ML. Psychological adjustment of children and adolescents with chronic arthritis: a meta-analytic review. *J Pediatr Psychol*. 1995;28(1):29-39. doi:10.1093/jpepsy/28.1.29.
10. Prchal A, Landolt M a. Psychological interventions with siblings of pediatric cancer patients: a systematic review. *Psychooncology*. 2009;18(12):1241-51. doi:10.1002/pon.1565.
11. Sharpe D, Rossiter L. Siblings of children with a chronic illness: a meta-analysis. *J Pediatr Psychol*. 2002;27(8):699-710. doi:10.1093/jpepsy/27.8.699.
12. Vermaes IPR, van Susante AMJ, van Bakel HJ. Psychological functioning of siblings in families of children with chronic health conditions: a meta-analysis. *J Pediatr Psychol*. 2012;37(2):166-84. doi:10.1093/jpepsy/jsr081.
13. Jutras S. Conception of Wellness in Families with a Diabetic Child. *J Health Psychol*. 2003;8(5):573-586. doi:10.1177/13591053030085008.
14. Knafl K, Breitmayer B, Gallo A, Zoeller L. Family response to childhood chronic illness: description of management styles. *J Pediatr Nurs*. 1996;11(5):315-26. doi:10.1016/S0882-5963(05)80065-X.
15. Lehna C. Fathers' and siblings' roles in families with children in home hospice care. *J Hosp Palliat Nurs*. 2001;3(1):1522-2179.
16. Loos M, Kelly S. Social Well-Being of Siblings Living with a Child with Diabetes. *Soc Work Health Care*. 2008;43(4):53-69. doi:10.1300/J010v43n04.
17. Kazak A, Segal-Andrews A, Johnson K. Paediatric psychology research and practice: a family systems approach. In: Roberts M, ed. *Handbook of Pediatric Psychology*. 2nd ed. New York: Guildford Press; 1995:84-104.
18. Cuskelly M. Adjustment of siblings of children with a disability: methodological issues. *Int J Adv Couns*. 1999;21:111-124.

19. Foster C, Eiser C, Oades P, et al. Treatment demands and differential treatment of patients with cystic fibrosis and their siblings: patient, parent and sibling accounts. *Child Care Health Dev.* 2001;27(4):349-64. doi:10.1046/j.1365-2214.2001.00196.x.
20. Gorodzinsky AY, Davies WH, Tran ST, et al. Adolescents' perceptions of family dynamics when a sibling has chronic pain. *Child Heal Care.* 2013;42(4):333-352. doi:10.1080/02739615.2013.842460.
21. James C, Holtzman N, Hadley DW. Perceptions of reproductive risk and carrier testing among adolescent sisters of males with chronic granulomatous disease. *Am J Med Genet C Semin Med Genet.* 2003;119C(1):60-9. doi:10.1002/ajmg.c.10007.
22. Alderfer M, Long K, Lown EA, et al. Psychosocial adjustment of siblings of children with cancer: a systematic review. *Psychooncology.* 2010;19(8):789-805. doi:10.1002/pon.1638.
23. Wilkins KL, Woodgate RL. A review of qualitative research on the childhood cancer experience from the perspective of siblings: a need to give them a voice. *J Pediatr Oncol Nurs.* 2005;22(6):305-319. doi:10.1177/1043454205278035.
24. Murray J. Siblings of children with cancer: a review of the literature. *J Pediatr Oncol Nurs.* 1999;16(1):25-34. doi:10.1177/104345429901600104.
25. MacLeod KD, Whitsett SF, Mash EJ, Pelletier W. Pediatric sibling donors of successful and unsuccessful hematopoietic stem cell transplants (HSCT): a qualitative study of their psychosocial experience. *J Pediatr Psychol.* 2003;28(4):223-30. Available at: <http://www.ncbi.nlm.nih.gov/pubmed/12730279>.
26. Critical Appraisal Skills Programme. CASP: making sense of evidence. <http://www.casp-uk.net/casp-tools>. Accessed November 2, 2016.
27. Thomas J, Harden A. Methods for the thematic synthesis of qualitative research in systematic reviews. *BMC Med Res Methodol.* 2008;8:45. doi:1471-2288-8-45 [pii]r10.1186/1471-2288-8-45 [doi].
28. Bellin MH, Kovacs PJ, Sawin KJ. Risk and protective influences in the lives of siblings of youths with spina bifida. *Health Soc Work.* 2008;33(3):199-209. doi:10.1093/hsw/33.3.199.
29. Wennick A, Huus K. What it is like being a sibling of a child newly diagnosed with type 1 diabetes: an interview study. *Eur Diabetes Nurs.* 2012;9(3):88-92. doi:10.1002/edn.213.
30. Read J, Kinali M, Muntoni F, Weaver T, Garralda ME. Siblings of young people with Duchenne muscular dystrophy-a qualitative study of impact and coping. *Eur J Paediatr Neurol.* 2011;15(1):21-8. doi:10.1016/j.ejpn.2010.07.006.
31. Herrman JW. Siblings' perceptions of the costs and rewards of diabetes and its treatment. *J Pediatr Nurs.* 2010;25(5):428-37. doi:10.1016/j.pedn.2009.07.004.
32. Derouin D, Jessee P. Impact of a chronic illness in childhood: siblings' perceptions. *Compr Pediatr Nurs.* 1996;19(2):135-147. doi:10.3109/01460869609038053.
33. Gallo A, Breitmayer B, Knafl K, Zoeller L. Stigma in childhood chronic illness: a well sibling perspective. *Pediatr Nurs.* 1991;17(1):21-25. Available at: <http://www.ncbi.nlm.nih.gov/pubmed/2000237>.
34. Hollidge C. Psychological adjustment of siblings to a child with diabetes. *Health Soc Work.* 2001;26(1):15-25. doi:10.1093/hsw/26.1.15.
35. Hutson SP. Experiences of siblings of patients with fanconi anemia. *Pediatr Blood Cancer.* 2007;48(1):72-79. doi:10.1002/pbc.20913.
36. Okashah R, Schoch K, Hooper SR, Shashi V, Callanan N. Parental communication and experiences and knowledge of adolescent siblings of children with 22q11.2 deletion syndrome. *J Genet Couns.* 2015;24:752-759. doi:10.1007/s10897-014-9806-4.

37. Velleman S, Collin SM, Beasant L, Crawley E. Psychological wellbeing and quality-of-life among siblings of paediatric CFS / ME patients : A mixed-methods study. *Clin Child Psychol Psychiatry*. 2015;1-16. doi:10.1177/1359104515602373.
38. Malcolm C, Gibson F, Adams S, Anderson G, Forbat L. A relational understanding of sibling experiences of children with rare life-limiting conditions: findings from a qualitative study. *J Child Health Care*. 2013. doi:10.1177/1367493513485825.
39. Altschuler J. *Working with Chronic Illness: a family approach*. London: Palgrave Macmillan; 1997.
40. Brennan C, Hugh-Jones S, Aldridge J. Paediatric life-limiting conditions: coping and adjustment in siblings. *J Health Psychol*. 2013;18(6):813-24. doi:10.1177/1359105312456324.
41. Stewart D, Stein A, Forrest G, Clark D. Psychosocial adjustment in siblings of children with chronic life-threatening illness: a research note. *J child Psychol psychiatry*. 1992;33(4):779-784. doi:10.1111/j.1469-7610.1992.tb00913.x.
42. Williams PD, Williams AR, Graff JC, et al. Interrelationships among variables affecting siblings and mothers in families of children with a chronic illness or disability. *J Behav Med*. 2002;25(5):411-424. doi:10.1023/A:1020401122858.
43. Compas BEB, Jaser SS, Dunn MJ, Rodriguez EM. Coping with chronic illness in childhood and adolescence. *Annu Rev Clin Psychol*. 2012;8:455-480. doi:10.1146/annurev-clinpsy-032511-143108.Coping.
44. Edwards M, Titman P. *Promoting Psychological Well-being in Children with Acute and Chronic Illness*. London: Jessica Kingsley Publishers; 2010.
45. Horowitz W, Kazak A. Family adaptation to childhood cancer: siblings and family system variables. *J Clin Child Psychol*. 1990;19(3):221-228. doi:10.1207/s15374424jccp1903_4.
46. Walker C. Stress and coping in siblings of childhood cancer patients. *Nurs Res*. 1988;37(4):208-212.
47. Sloper P. Experiences and support needs of siblings of children with cancer. *Health Soc Care Community*. 2000;8(5):298-306. doi:10.1046/j.1365-2524.2000.00254.x.
48. Wilkins KL, Woodgate RL. A review of qualitative research on the childhood cancer experience from the perspective of siblings: a need to give them a voice. *J Pediatr Oncol Nurs*. 2005;22(6):305-19. doi: 10.1177/1043454205278035.
49. Alderfer MA, Long KA, Lown EA, Marsland AL, Ostrowski NL, Hock JM, Ewing LJ. Psychosocial adjustment of siblings of children with cancer: a systematic review. *Psychooncology*. 2010;19(8):789-805. doi: 10.1002/pon.1638.
50. Long KA, Marsland AL. Family adjustment to childhood cancer: a systematic review. *Clinical Child and Family Psychology Review*. 2011;14(1):57-88. doi: 10.1007/s10567-010-0082-z.
51. Drotar D, Crawford P. Psychological adaptation of siblings of chronically ill children: research and practice implications. *Dev Behav Pediatr*. 1985;6(6):355-362.
52. Nielsen KM, Mandleco B, Roper SO, Cox A, Dyches T, Marshall ES. Parental perceptions of sibling relationships in families rearing a child with a chronic condition. *J Pediatr Nurs*. 2012;27(1):34-43. doi:10.1016/j.pedn.2010.08.009.

Tables and figures

Table 1 Search Terms Used for Meta-synthesis

Search terms	Boolean terms: 'OR' used within row and 'AND' used across rows
Sibling	brother, sister, kin
Experience	experien*, cop*, resilien*, manage*, deal*, respon*, adapt*, adjust, understand*, impact, well-being, wellbeing, effect*, thought, think, feel, perspective
Chronic non-communicable physical health condition	chronic health condition, chronic physical health condition, chronic disease, Muscular Dystrohy, Cancer, neoplasm, Asthma, Cystic Fibrosis, Diabetes, Arthritis, life-limiting, fatal, terminal, congenital, genetic, non-communicable, organic disease, non-infectious disease, autoimmune disease, kidney disease, heritable, patient, palliative
Qualitative	qualitative, interview, IPA, grounded theory, subjective, quot*, narrative*, them*, audio, discourse, phenomenon*
Not to be included	autism, ASC, psychosis, psychotic, schizophrenia, bipolar, Alzheimer, attention deficit disorder, ADHD, anorexia, bulimia, HIV, drug

Figure 1 PRIMSA Flow Chart

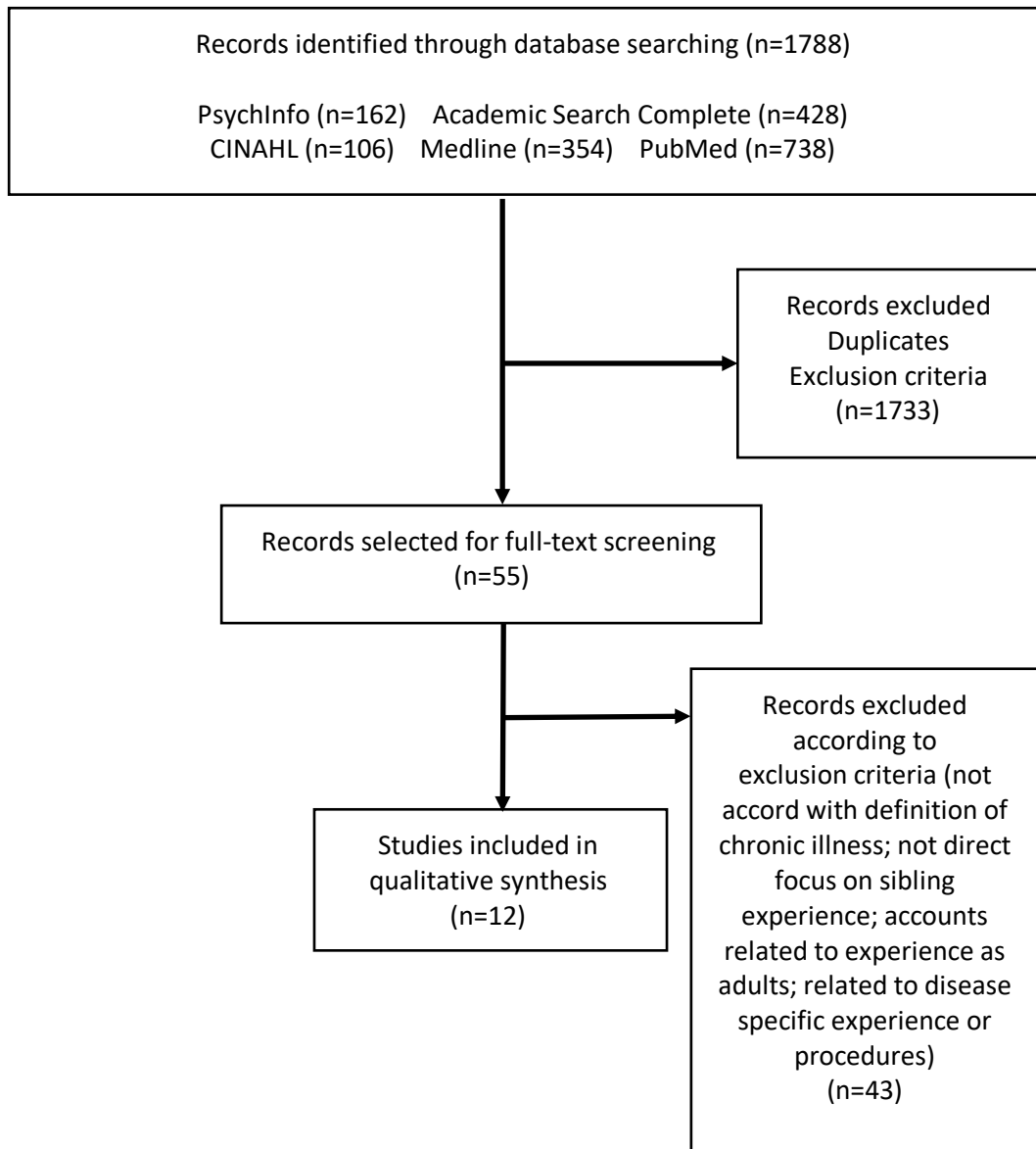


Figure 2 Themes and connections

Theme 1: Changing relationships

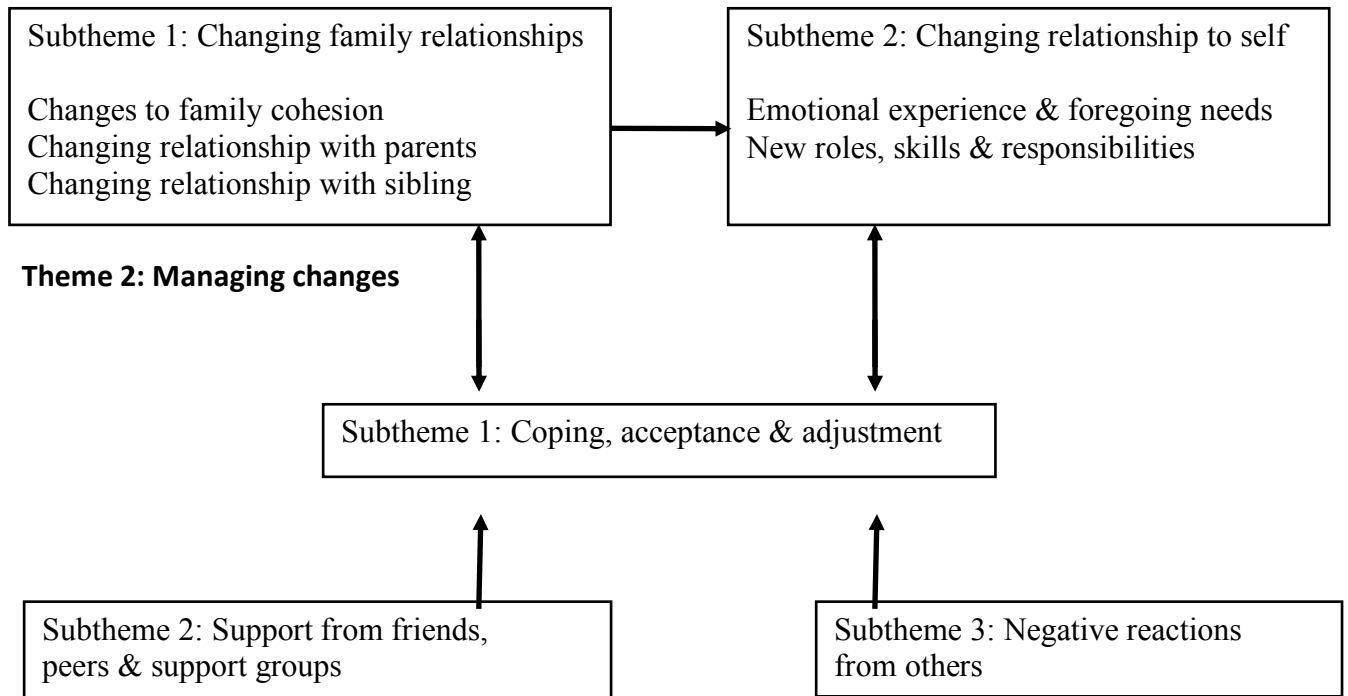


Table 2 Summary of Key Features of Selected Articles

Author/Date	Title	Methodology	Participants	Age range or average of healthy siblings (unwell siblings)	Type of illness	Setting and country
Bellin et al (2008) ²⁸	Risk and protective influences in the lives of siblings of youths with spina bifida	Data reported from larger mixed method study. One open ended question was posed in posted study packet; written responses returned by post. Thematic content analysis.	155 siblings	13.83	Spina Bifida	Social work, USA
Brennan et al (2012) ⁴⁰	Paediatric life-limiting conditions: coping and adjustment in siblings	Mixed methods, semi-structured interviews. Grounded theory.	31 siblings	9.9	Life-limiting conditions	Health Psychology, UK
Derouin & Jessee (1996) ³²	Impact of a chronic illness in childhood: siblings' perceptions	Semi-structured telephone interviews and questionnaires to siblings and parents.	15 Siblings 14 parents	8-17	Chronic illnesses	USA
Gallo et al (1991) ³³	Stigma in childhood chronic illness: a well sibling perspective	Part of a larger study. Structured interview with results categorised according to questions.	27 Siblings	6-14	Chronic illnesses	Nursing, USA
Herrman (2010) ³¹	Siblings' perceptions of the costs and rewards of diabetes and its treatment.	Semi-structured interviews focusing on rewards and costs of having a sibling with diabetes, developed according to social exchange theory. Template analysis.	20 Siblings	4-16	Diabetes	Nursing, USA
Hollidge (2001) ³⁴	Psychological adjustment of siblings to a child with diabetes.	Mixed methods/Semi-structured interviews. References not given for qualitative part of analysis.	28 Siblings	8-12	Diabetes	Social Work, Canada
Hutson & Alter (2007) ³⁵	Experiences of siblings of patients with fanconi anemia	Semi-structured interviews. Reported using qualitative description and content analysis.	9 siblings	11-21	Fanconi Anemia	Nursing, USA
Malcolm et al (2013) ³⁸	A relational understanding of sibling experiences of children with rare life-limiting conditions: Findings from a qualitative study	Mixed methods study. Qualitative section used interviews facilitated by card sort technique. Analysis informed by grounded theory.	8 siblings	7-12	Mucopolysacchari doses & Batten disease	Nursing, UK
Okashah et al (2015) ³⁶	Parental communication and experiences and knowledge of	Mixed methods, online survey with 2 open ended questions. Reported using	29 siblings	12-16	22q11.2 deletion syndrome	Genetic counselling,

	adolescent siblings of children with 22q11.2 deletion syndrome	content analysis.				USA
Read et al (2011) ³⁰	Siblings of young people with DMD - a qualitative study of impact and coping	Semi-structured interviews using thematic analysis and the thematic framework approach for analysis.	35 siblings	11-18 (5-22)	Duchenne Muscular Dystrophy	Neurology, UK
Velleman et al (2015) ³⁷	Psychological wellbeing and quality of life among siblings of paediatric CFS/ME patients: A mixed-methods study	Mixed methods, semi-structured interviews and analysed using thematic analysis.	9 siblings	12-17 (8-18, 13.8 mean)	CFS/ME	Clinical Psychology/Psychiatry, UK
Wennick & Huus (2012) ²⁹	What it is like being a sibling of a child newly diagnosed with type 1 diabetes: an interview study	Semi-structured interviews and content analysis.	7 siblings	10-17 (8-13, 9.5 median)	Diabetes	Nursing, Sweden

Table 3 CASP Quality Appraisal

Articles	Presence of clear research aims?	Qualitative approach appropriate?	Design appropriate for aims?	Recruitment strategy appropriate?	Does data collection address the research issue?	Presence of reflection on researcher and participant relationship?	Ethical issues considered?	Data analysis rigorous?	Clear statement of findings?	Does research add value?	Total score out of 30
Bellin et al (2008) ²⁸	3	2.5	1.5	1.5	2	1	1.5	2.5	2.5	3	21
Brennan et al (2012) ⁴⁰	3	2.5	3	2	3	0	2	2.5	2.5	2	22.5
Derouin & Jessee (1996) ³²	3	2.5	1.5	2	1.5	1	1.5	1	2	2	18
Gallo et al (1991) ³³	3	2.5	1	1	2	0	0	1	1.5	1.5	13.5
Herrman (2010) ³¹	3	2.5	3	2	3	0.5	1.5	2	2	2	21.5
Hollidge (2001) ³⁴	2.5	2.5	1.5	2	1	0	0	1	1.5	2	14
Hutson & Alter (2007) ³⁵	2.5	2.5	3	2.5	3	1	2.5	2	2	2.5	23.5
Malcolm et al (2013) ³⁸	3	2.5	3	2.5	2.5	0.5	2	1.5	2.5	1.5	21.5
Okashah et al (2015) ³⁶	2.5	2	1	1.5	1.5	1	1	1	1	1	13
Read et al (2011) ³⁰	3	2.5	2	2	2.5	0.5	2	2	2.5	1.5	20.5
Velleman et al (2015) ³⁷	3	2	1.5	2	1.5	1	1.5	1.5	2	2	17.5
Wennick & Huus (2012) ²⁹	2.5	2.5	2.5	2	2	0.5	2.5	1.5	2	1.5	19.5