The self-reported experiences of siblings who have a brother or sister diagnosed with childhood cancer: a systematic review protocol of qualitative evidence

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Review question/objective

The overall objective of this review is to promote a broader understanding of the experiences of siblings of a brother or sister diagnosed with childhood cancer. The specific objectives include to synthesize existing findings and to summarize the best available evidence to increase the overall understanding of what it is like to live with a brother or sister with cancer. The recommendations derived from this study may provide further information for clinical nursing to understand the needs of
siblings who have a brother or sister diagnosed with childhood cancer, to offer evidence based recommendations toward providing appropriate care and support for these siblings of children with cancer, to promote their adaptation and mental health, and to provide the families of children with cancer care reference, to promote a family-centered care model.

The review questions are:

1. What are the experiences of siblings who have a brother or sister diagnosed with a childhood cancer?

2. What are the experiences of siblings who have a brother or sister diagnosed with a childhood cancer regarding the impact on their lives?

**Background**

Cancer is the second most common cause of death, after accidents, in childhood and a significant public health problem in developed countries. Owing to highly specific diagnostic procedures and the introduction and continuous improvement of multimodal treatment strategies, the past decades have led to dramatic improvements in survival rates for most types of childhood cancer with a marked rise in the probability of cure.

Childhood cancer is a highly stressful experience that can challenge and disrupt the family dynamic and its members. As cancer is a life threatening illness, when a child is diagnosed with cancer the whole family is affected. With the need to care for an ill child, a family is forced to change the normal daily life routines, whilst also trying to maintain a quasi-normal way of life. Many of the family stresses and changes accompanying childhood cancer have a severe impact on siblings.

The family is viewed as an interactional system. Family members are connected to each other, exhibit coherent behaviors, and share some degree of interdependence. Families are in constant interaction with their physical, social, and cultural environment. A layering of subsystems is found within family systems, such as the spouse subsystem, the parent-child subsystem, the sibling subsystem, etc. Therefore the family is a complex, integrated whole. Within a family system, patterns of interdependence result in all members of the family system being impacted by change in one another.

When a child in the family is stricken with a serious disease, almost every family enters into a crisis situation. The family must adapt to accommodate the ill child's needs. Thus, the family system must be dynamic, and there is often interdependence among its members. The sibling subsystem is an important family subsystem in which a child and his or her brothers and sisters interact as they grow up. The relationships of siblings serve as the first social-skill learning for children. Normally the sibling relationship helps a child learn how to share, support, become angry, negotiate, compromise, compete, cooperate, and imitate each other. The closeness that develops among them eventually leads to a very special, life-long relationship. However, when one child becomes ill, the other healthy siblings may suffer with confused feelings, as the relationship between them and the ill child changes.

When parents focus their attention on the child diagnosed with cancer, the parents may neglect the other child at home and healthy siblings may be confronted with decreased physical and emotional availability of their parents. The impact on the healthy sibling/s include: (a) change in psychological status, i.e., increase in worry about life, anxiety about the disease, and negative self-concepts (b)
change in family interaction, i.e., relationship with mother, relationship with father, relationship with other siblings (c) change in social relationship, i.e. hunger for companions, changing recreation.

Prchal and Landolt conducted a qualitative study to determine the experiences of siblings of pediatric cancer patients in different areas of life in the first half-year after the cancer diagnosis. They investigated five areas of the siblings' daily life: hospital, school, peers, family life, and ill child. Siblings were very much aware of the consequences of the illness and the treatment for the ill child. They observed their brothers' or sisters' appearance, suffering, behavior change, and their being the center of attention. Siblings described how family life after the cancer diagnosis was determined by the ill child's treatment protocol, resulting in their parents' regular absence and reduced family activities. Living together with a sick brother or sister also resulted in sibling jealousy and evoked intrusive worries about the ill child's health and prognosis. Interviewed siblings also expressed worries about their brother's or sister's altered behavior, and it was difficult for siblings to observe the ill child's changed appearance.

If the child was hospitalized, siblings also reported being affected by other patients in the hospital. Witnessing other ill children suffering, observing their altered appearance, or even hearing about the death of a patient was difficult for siblings. Further difficulties emerged at school, with siblings reporting a decline in academic performance, and an aversion to taking part in everyday life during the time they felt the need to support the ill child. Siblings' peer activities were reduced and difficult to maintain because of the changed situation in the family. However, not all the experiences of siblings were negative. Siblings' accounts indicated that supportive relationships with peers were resources that helped them cope with the situation. Siblings also reported increased closeness and a strengthening of bonds between family members after the experience of acute illness in the family. This finding of positive experiences is consistent with results of earlier studies.

In a study by Iles, five healthy school age siblings of children with cancer were interviewed. The findings indicated themes which included the change of interpersonal relationships, the change of self concept, and loss and growth. The loss experience of healthy siblings included disruption of interpersonal relationships (especially with parents, siblings and peers), physical dysfunctions, and disturbances in the routine of family life. These circumstances provided opportunities for the emotional growth of the siblings involved. Siblings perceived some positive effects of the cancer experience, such as increased empathy for parents needs.

A literature review by Wilkins and Woodgate on the childhood cancer experience from the perspective of siblings examined 27 articles from 1979 to 2004. In this study, the most frequently used research design was descriptive exploratory. The sampling methods included convenience, theoretical purposive, and random. Most studies employed interviews for data collection. The most commonly used qualitative data analysis procedures were the constant comparative method and content analysis. The number of sibling participants across studies ranged from one to 254. Sixty-three percent of the studies included both brothers and sisters. Siblings' ages ranged from five to 40 years, with the majority between the ages of seven and 18 years. The findings were categorized into three themes: (a) Siblings perceived tremendous change in family relationships and their family dynamics and routines, including losses (e.g. losses of parental attention, normal family roles, sense of self) and gains (e.g. increased family closeness, independence, maturity, empathy); (b) intense feelings (e.g. sadness, loneliness, rejection, anxiety, anger, jealousy, guilt), and (c) unmet sibling needs (e.g. needs for open and honest communication, more information and involvement in the treatment process, and greater support.
maintaining personal interests and activities). These findings highlight the fact that siblings missed their own and their family's usual activities and routines, and they often felt neglected. It also highlighted that it was important for siblings to receive information about the ill child's cancer and treatment. Furthermore, siblings perceived intense feelings; these results, like other studies, have demonstrated that siblings of children with cancer experience negative emotional reactions in themselves and family members, such as worry about their ill brother or sister, increased sibling rivalry, anger and frustration, rejection and guilt; feelings of hate, jealousy and isolation.

To understand the experience of a sibling who has a brother or sister diagnosed with cancer, it is important to allow them to tell the stories of their experience in their own words. Approaching the study of siblings' experiences from the qualitative research paradigm provides valuable insights into the changes siblings encounter, the intensity of their feelings and the resources that they find helpful. Qualitative meta-synthesis is a useful method for bringing together studies that explore the same or closely related phenomena. The goals of qualitative meta-synthesis are theory development, higher level abstraction, and generalizability in order to make qualitative findings more accessible for application in practice. Qualitative meta-synthesis increases the usefulness of constituent study findings by creating more comprehensive views of phenomena. Although there is qualitative research available that discusses the experience of siblings of children with cancer, synthesizing the data into a systematic review may provide a better understanding of the overall experiences of siblings of children with cancer. This may facilitate the development of knowledge that would be useful to pediatric nurses and those involved in care of the children. Furthermore, knowledge about siblings' perspective of the childhood cancer may provide insight into the experience of the family system. The study's findings may highlight the philosophy that nursing care needs to be family-centered.

An initial search of CINAHL, MEDLINE/PubMed, Eric, and The Joanna Briggs Institute Library of Systematic Reviews was conducted to identify any previous systematic review on this topic. There was a systematic literature review by Wilkins and Woodgate on the childhood cancer experience from the perspective of siblings. The review focused on the contributions of qualitative research in advancing our understanding of the childhood cancer experience from the perspective of siblings. The focus of the research was to review qualitative studies on the broad topic of siblings of children with cancer, siblings' ages ranged from five to 40 years. However, the present review differs from Wilkins and Woodgate in several significant ways. Firstly, the review aims to promote overall understanding of the experience of siblings of a brother or sister diagnosed with childhood cancer. Secondly, the review will be conducted on the experiences of siblings aged from six to 20 years, who have a brother or sister diagnosed with a childhood cancer and their voice that describes the experience. Thirdly, this review aims to cover a wider range of databases and cultures than the previous review, and to review literature in both the Chinese and English languages.

**Keywords**

child*; children; childhood; adolescen*; teen*; youth*; pediatric; paediatric; cancer; neoplasm; carcinoma; sarcoma; lymphoma; neuroblastoma; tumor; leukemia; brain tumor; siblings; sister; brother; family; lived experience*; life experience; feeling*; child parent relation; psychosocial adjustment; distress; stress; impact
Inclusion criteria

Types of participants

This review will consider studies that include the siblings aged from six to 20 years, who have a brother or sister diagnosed with childhood cancer. Studies will be considered for inclusion, that have siblings of children with cancer as the sole research participants or as research participants within the context of the family. There will be no restriction on the type, severity, and prognosis of cancers.

Phenomena of interest

This review will consider studies that investigate the self-reported experiences of siblings of children with cancer, or the impacts on the siblings of children with cancer.

Context

The settings will include within the home, clinical setting, and/or community setting of any cultural context.

Types of studies

This review will consider studies that focus on qualitative data including, but not limited to, designs such as qualitative research, phenomenology, hermeneutic phenomenology, grounded theory, ethnography, action research, focus group and narrative research. In the absence of research studies, expert opinion papers and reports will be considered in a narrative summary.

Search strategy

The search strategy aims to find both published and unpublished studies. A three-step search strategy will be utilized in this review. An initial limited search of MEDLINE and CINAHL will be undertaken followed by analysis of the text words contained in the title and abstract, and of the index terms used to describe an article. A second search using all identified keywords and index terms will then be undertaken across all included databases. Thirdly, the reference list of all identified reports and articles will be searched for additional studies. Studies published in the English or Chinese language will be considered for inclusion in this review. Studies published from 1960 to 2013 will be considered for inclusion in this review.

The databases to be searched include:

CINAHL
MEDLINE/PubMed
Science Direct
Scopus
Eric
JBI Library
The databases searched for articles written in Chinese will include:

- The electronic theses dissertations system
- Chinese electronic periodical services
- Airiti Library
- Taiwan academic online
- Google scholar

The search for unpublished studies will include:

- The electronic theses dissertations system

Initial keywords to be used will be:

**English search terms**

a. Types of participants: child* or children or childhood or adolescen* or teen* or youth* or pediatric or paediatric or cancer or neoplasm or carcinoma or sarcoma or lymphoma or neuroblastoma or tumor or leukemia or brain tumor or siblings or sister or brother or family

b. Phenomena of interest: lived experience* or life experience or experiences or feeling* or child parent relation or psychosocial adjustment or distress or stress or impact

c. Types of studies: qualitative research or phenomenology or hermeneutic phenomenology or grounded theory or ethnography or focus group or action research or narrative research

**Chinese search terms**

a. Types of participants: 癌症、腫瘤、兒童、青少年、手足、家庭

b. Phenomena of interest: 經驗、親子關係、心理社會適應、壓力

c. Types of studies: 質性研究、現象學、詮釋現象學、紮根理論、民族誌、焦點團體、行動研究、敘事研究

**Assessment of methodological quality**

Qualitative papers selected for retrieval will be assessed by two independent reviewers for methodological validity prior to inclusion in the review using standardized critical appraisal instruments from the Joanna Briggs Institute Qualitative Assessment and Review Instrument (JBI-QARI) (Appendix I). Any disagreements that arise between the reviewers will be resolved through discussion, or with a third reviewer.
Textual papers selected for retrieval will be assessed by two independent reviewers for authenticity prior to inclusion in the review using standardized critical appraisal instruments from the Joanna Briggs Institute Narrative, Opinion and Text Assessment and Review Instrument (JBI-NOTARI) (Appendix I). Any disagreements that arise between the reviewers will be resolved through discussion, or with a third reviewer.

**Data collection**

Qualitative data will be extracted from papers included in the review using the standardized data extraction tool from the JBI-QARI (Appendix II). The data extracted will include specific details about the phenomena of interest, populations, study methods and outcomes of significance to the review question and specific objectives.

Textual data will be extracted from papers included in the review using the standardized data extraction tool from JBI-NOTARI (Appendix II). The data extracted will include specific details about the interventions, populations, study methods and outcomes of significance to the review question and specific objectives.

**Data synthesis**

Qualitative research findings will, where possible be pooled using the JBI-QARI. This will involve the aggregation or synthesis of findings to generate a set of statements that represent that aggregation, through assembling the findings (Level 1 findings) rated according to their quality, and categorizing these findings on the basis of similarity in meaning (Level 2 findings). Finally a meta-synthesis will be carried out to produce a single comprehensive set of findings (Level 3 findings) that can be used as a basis for evidence-based practice. Where textual pooling is not possible the findings will be presented in narrative form.

Textual papers will, where possible be pooled using JBI-NOTARI. This will involve the aggregation or synthesis of conclusions to generate a set of statements that represent that aggregation, through assembling and categorizing these conclusions on the basis of similarity in meaning. These categories are then subjected to a meta-synthesis in order to produce a single comprehensive set of synthesized findings that can be used as a basis for evidence-based practice. Where textual pooling is not possible the conclusions will be presented in narrative form.

**Conflicts of interest**

No potential conflict of interest noted.

**Acknowledgements**

The authors acknowledge Taiwan Joanna Briggs Institute Collaborating Center and Taiwan Evidence-Based Nursing Association for their support in this review. We also acknowledge The Joanna Briggs Institute and Dr Matthew Stephenson for his help in the peer review process.
References


Appendix I: Appraisal instruments  QARI Appraisal instrument

**JBI QARI Critical Appraisal Checklist for Interpretive & Critical Research**

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<th>Question</th>
<th>Yes</th>
<th>No</th>
<th>Unclear</th>
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<td>1. Is there congruity between the stated philosophical perspective and the research methodology?</td>
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<td>4. Is there congruity between the research methodology and the representation and analysis of data?</td>
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<td>6. Is there a statement locating the researcher culturally or theoretically?</td>
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<td>7. Is the influence of the researcher on the research, and vice versa, addressed?</td>
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<td>8. Are participants, and their voices, adequately represented?</td>
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<td>9. Is the research ethical according to current criteria or, for recent studies, and is there evidence of ethical approval by an appropriate body?</td>
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Overall appraisal: □ Include □ Exclude □ Seek further info. □

Comments (including reason for exclusion)

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NOTARI Appraisal instrument

**JBI Critical Appraisal Checklist for Narrative, Expert opinion & text**

Reviewer ........................................ Date ........................................

Author ........................................ Year ....... Record Number ...........

1. Is the source of the opinion clearly identified? [ ] Yes [ ] No [ ] Unclear [ ] Not Applicable
2. Does the source of the opinion have standing in the field of expertise? [ ] Yes [ ] No [ ] Unclear [ ] Not Applicable
3. Are the interests of patients/clients the central focus of the opinion? [ ] Yes [ ] No [ ] Unclear [ ] Not Applicable
4. Is the opinion's basis in logic/experience clearly argued? [ ] Yes [ ] No [ ] Unclear [ ] Not Applicable
5. Is the argument developed analytically? [ ] Yes [ ] No [ ] Unclear [ ] Not Applicable
6. Is there reference to the extant literature/evidence and any incongruency with it logically defended? [ ] Yes [ ] No [ ] Unclear [ ] Not Applicable
7. Is the opinion supported by peers? [ ] Yes [ ] No [ ] Unclear [ ] Not Applicable

Overall appraisal: Include [ ] Exclude [ ] Seek further info [ ]

Comments (including reason for exclusion)

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Appendix II: Data extraction instruments

QARI data extraction instrument

**JBI QARI Data Extraction Form for Interpretive & Critical Research**

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<td>Year</td>
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<td>Journal</td>
<td>Record Number</td>
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**Study Description**

- Methodology
- Method
- Phenomena of interest
- Setting
- Geographical
- Cultural
- Participants
- Data analysis
- Authors Conclusions
- Comments

Complete: Yes ☐ No ☐
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<td>Extraction of findings complete</td>
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# NOTARI data extraction instrument

## JBI Data Extraction for Narrative, Expert opinion & text

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### Study Description

**Type of Text:**

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**Those Represented:**

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**Stated Allegiance/ Position:**

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**Setting**

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**Cultural**

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**Logic of Argument**

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**Data analysis**

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**Authors Conclusions**

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**Reviewers Comments**

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**Data Extraction Complete**

Yes ☐

No ☐
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<th>Conclusions</th>
<th>Illustration from Publication (page number)</th>
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Include:  
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