

# Functional limitations caused by simple bone cysts

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## Abstract

**Purpose** Relatively little is known about the impact of benign bone lesions on function. The aim of this study was to create a more complete understanding of the impact of functional disability from simple bone cysts (SBCs) by combining qualitative and quantitative methods.

**Methods** This study followed a convergent parallel mixed methods design. The quantitative arm included 130 children with SBC and used the Activities Scale for Kids (ASK) to measure physical function. In the qualitative arm ten children and their parents participated in interviews related to activity participation and interactions with their physical and social environments. The two data sets were analyzed independently and then the results were integrated.

**Results** The ASK demonstrated 35% of children achieving the maximum score. In total, 65% of children responded “I had no medical needs” confirming that SBC, while being present throughout childhood, is largely perceived as not a chronic illness. Qualitatively most children reported minimal or no changes in activity participation but reported thinking about being more cautious during play, confirming that SBC affects effort not participation in play.

**Conclusion** The diagnosis of SBC did not have a significant impact on physical function, but did alter children's thoughts

about physical activity participation. This finding suggests that physical function scores may have unappreciated ceiling effects. Outcome tools that combine both illness perceptions and physical function may help to better assess functional outcomes of SBC.

*Level of evidence:* III

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**Keywords:** bone cysts; outcomes; mixed methods

## Introduction

The main goal of simple bone cyst (SBC) treatment is to reduce the risk of pathological fracture and allow children to return to normal activity.<sup>1</sup> Although several studies have reported on functional outcomes following SBC, none have studied both function and the psychological impact of the diagnosis related to activity participation of children and their families.<sup>2-4</sup> The perceptions of children and families regarding SBC diagnosis, treatment and outcomes, therefore, remain poorly understood.

Several approaches can be used to understand the impact of SBCs. Qualitative studies have been increasingly used across orthopaedics over recent years and can be used to better understand patient and family perceptions and the experience of living with a SBC.<sup>5-7</sup> Quantitative studies can be used to understand the clinical and functional outcomes of SBC. The aim of this study was to create a more comprehensive understanding of the impact of functional disability and changes related to SBC through combining both qualitative and quantitative methods in a mixed methods design.

## Patients and methods

This study followed a convergent parallel, mixed methods design to investigate the effects and impact of SBCs on children and their families.<sup>8</sup> The main question, ‘what are the impact and functional limitations caused by SBC?’, was analyzed by both qualitative and quantitative methods, which were completed independently. The results of these two arms were then combined to provide a more comprehensive understanding. Neither arm of the study functioned as a control arm but rather results from both data sets were integrated to achieve the aim of the study.

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### Recruitment

The quantitative data used in this study was collected as part of a randomized clinical trial comparing two interventions (intralesional bone marrow or steroid injection) for the management of SBCs in 130 patients.<sup>9</sup> The study found no differences regarding functional outcomes between treatments groups and the patients from study were, therefore, considered a single group for the purposes of this study. Eligibility criteria included children diagnosed with SBC < 18 years of age and excluded children with an underlying musculoskeletal condition such as osteogenesis imperfecta, malignancy or chronic steroid use. The same criteria were maintained for recruitment of patients for the qualitative arm of this study. All patients who participated in a qualitative interview were given a \$10 gift card in appreciation of their participation. This study received approval from our institution's Children Research Ethics Board and all procedures performed involving human participants were in accordance with the ethical standards of the 1964 Helsinki declaration and its later amendments.

### Data collection

In the quantitative arm, physical function was assessed using the Activities Scale for Kids (ASK).(10) The ASK is a 30-item self-reported questionnaire developed in 1994 as a generic tool for assessing physical function in children. The ASK has been shown to be reliable, validated and responsive.<sup>10-12</sup> Possible scores for the ASK range from 0 to 100 with higher scores indicating better function.<sup>12,13</sup> All children who were recruited for participation in interviews were also asked to complete the ASK following the interview.

### Qualitative data

The qualitative arm of this study followed the methodological tradition of phenomenology to study the lived experience of children with SBCs.<sup>14,15</sup> Sample size was determined by reaching data saturation, a point defined by when review of additional interview transcripts produced no new codes or new concepts from children or parents.<sup>16,17</sup>

A semi-structured interview guide was developed through discussion with clinicians who had expertise in SBCs and review of literature. Interview questions focused on two main topics: a child's activity participation and interactions with their physical and social environments. Questions were broad and open-ended and then responses were probed to reach deeper and more specific feelings. Children and parents were interviewed separately in the child's home by a single interviewer. Interviews lasted between 30 and 45 minutes in total. All interviews were audio recorded and transcribed verbatim. A reflexive journal, memos and an audit trail were kept to record methodological and logistical decisions

during initial coding and subsequent analysis, as well as the values and interests of the researcher related to the study to increase transparency and reflexivity.<sup>18,19</sup> Reflexivity involves acknowledging that the researcher is part of the context in which a phenomenon is being studied and includes self-reflection regarding biases and theoretical dispositions.<sup>20</sup> This was reviewed prior to data analysis independently by the author (AB) and with the research team (CF, JW) in order to limit bias and improve validity of the research. The initial review of the qualitative data was conducted after the first few interviews and iterative adjustments were made to the interview guide.

### Data analysis

ASK scores and demographic data from the interview cohort were compared with the larger trial cohort. The postoperative children who were interviewed were near the one-year post-surgical benchmark and had had a final follow-up radiograph and clinical visit resulting in the removal of all activity restrictions. The one-year outcome data from the trial was used as the outcome measure as most of these children had also received clearance for return to all activity.

Braun and Clarke's<sup>21</sup> phases of thematic analysis were employed to analyze interview transcripts. A preliminary review of the transcripts was conducted to generate a list of provisional codes. Transcripts were coded using the MAXQDA software package (VERBI Software, 2019). Provisional coding was done by the primary researcher, and codes were then discussed with the research team and consensus was achieved prior to initial coding.<sup>18</sup> Codes were subsequently collated into candidate themes and then employed to review the entire data set for patterns.<sup>21</sup> Constant comparison was used to compare and contrast concepts within and across interviews.

### Integration

Qualitative and quantitative findings at the one-year outcome mark were integrated after independent analysis of each data set. Integrative analysis was conducted to identify if the results of the quantitative ASK questionnaire adequately captured the qualitative responses of study participants.<sup>14</sup>

## Results

### Quantitative

One-year ASK scores were available for 103 patients including 32 girls and 71 boys between three and 17 years of age. Of these, 73 children had a SBC affecting the upper extremity and 30 affecting the lower extremity. All patients were treated surgically. At one year 27% of cysts demonstrated radiographic healing and the mean cyst size

was 71.5 cm<sup>3</sup> compared with 79.4 cm<sup>3</sup> at baseline. A comparison of the demographics of the trial cohort and interviewed cohort is presented in Table 1 with chi-squared tables for sex and extremity and Wilcoxon-rank-sum tests for age and ASK scores showing no statistically significant differences between the two groups (p-value set at 0.05).

ASK scores ranged from 52 to 100 with a mean of 94.8 points. In all, 67 patients had ASK scores < 100. Each of the 30 items of the ASK were assessed. In total, 65% of patients replied: not applicable (checking the box labelled "I did not have special medical needs") to an item that asked specifically about medical needs.

### Qualitative

The qualitative findings revealed three main themes related to: activity participation; concern and anxiety related to activity; and the impact of SBC on parents and families.

#### Activity participation

Children, often presenting initially with pathological fracture, described the experience of SBC, particularly the activity restrictions and limitations, as: "just like having a broken bone". As one child put it, "Soon after [the sling came off] I had full mobility again, so it was really just like a broken arm" (boy, 17 years). This pattern of fracture followed by a short period of limitation and a subsequent quick return to activity was repeated even after children suffered multiple fractures due to the SBC. One parent described this experience saying, "It didn't impede our pattern of behaviour regarding activity after three breaks" (mother of boy, ten years). Despite repeated fractures and periods of limited activity, children did not describe long-term reductions in sport participation, or their functional abilities, due to the SBC. The illness (i.e. SBC) causing the fracture did not seem to be of great importance to children.

#### Concerns and thoughts about activity

While children did not stop participating in activities or sports due to their SBC, there was a sense of concern, particularly amongst those who had experienced fracture(s)

but had not yet had a surgical intervention. While only few used the word 'weak' to describe their limb, almost all described fear of re-injuring themselves while participating in physical activity. Children described concern or anxiety when thinking about participating more than when actually participating in activity. One child said "Sometimes I think about it when I'm by myself" (boy, ten years) and one parent noted that "I think sometimes, when it was quiet at night he thinks – 'I was playing dodgeball today...could I break it again?' - But during the day when he's doing most things, I don't really think he thinks about it" (father of boy, 17 years). Often the experience was described as a subtle thought that did not prevent them from playing but caused them to exercise a little more caution during play or to hesitate briefly before beginning participation. Pre-operative children tended to talk more about being cautious while playing sports but also emphasized that their level of participation and enthusiasm were unaffected. Postoperative children contrarily described remembering feelings of being cautious during play when they were preoperative but no longer felt the concerns and anxieties that resulted in cautious participation.

#### Family impact

The items on the ASK do not include questions that provide information specific to the impact of the condition on the family. Interview data provided an answer that was fairly uniform across families. Families of children with SBC maintained their regular activities, routines and vacations but often made special accommodations for the affected child. Some families skipped specific activities that the child would not be able to participate in but typically used this time for a similar alternative. For example, one family went to the pool where the child could swim instead of the jungle gym. In some cases, the child had to miss certain activities, such as waterskiing at the cottage in one case, but was still able to be involved by riding in the boat. Children seemed to understand and appreciate these accommodations as one child said, "Sometimes I would go out with my dad in the boat.... I didn't really care [about not waterskiing and tubing]" (boy, ten years).

Like children, parents whose children were preoperative seemed more concerned about their child's risk of re-fracture and exercised caution or attempted restriction of the child's activity. However, these restrictions did not seem to last beyond the short-term of several weeks following fracture healing and postoperative parents, who's children were around one year from completion of treatment, admitted that they no longer had concerns about their child participating in physical activity without restraint. Preoperative parents were more likely to enforce short-term restrictions with one parent stating, "Due to the fact that there was no cast he would do things thinking his arm

**Table 1** Demographic features of trial and interview cohorts

	Trial cohort	Interview cohort	p-value*
Number	103	10	
Sex			0.466
Girls	32	2	
Boys	71	8	
Extremity			0.542
Upper	73	7	
Lower	30	3	
Mean age, yrs (range)	9.6 (3 to 17)	9.5 (4 to 17)	0.785
Mean ASK score (sd)	95 (7.9)	90 (11.7)	0.114

\*Chi-square/t-test  
ASK, Activities Scale for Kids

was better, so just in that short period after a fracture, then I would restrict his activity" (mother of boy, ten years).

### Integration

There were similarities between the quantitative and qualitative results, with the qualitative results providing deeper insight into the more subtle effects of SBC on physical function. For example, the qualitative theme of children perceiving SBC as "just a broken bone" and the quantitative analysis of the ASK items showing overall very good function. Additionally, the qualitative finding that children perceived themselves simply as injured may explain the high rate of the "not applicable" response to ASK items related to medical needs.

## Discussion

This study demonstrated that overall the functional limitations of SBCs are minimal with a mean score of 94.8 points. Previous studies have established a mean ASK score of 96 in children with no abnormality and the patients in this study population had similar results.<sup>13</sup> Qualitative interviews also revealed that children's perceptions of their function were relatively unchanged from prior to the diagnosis.

Connors and Stalker<sup>22</sup> studied children's experiences of disability in 2003 and similarly found that children, even with significant disabilities, viewed their limitations as "not a big deal". Similarly Cartwright et al<sup>23</sup> found that children with juvenile idiopathic arthritis (JIA) did not "allow [JIA] to hold me back". Children with SBC not perceiving themselves as ill may have several sources. Based on the work of these previous authors, there may be some protective effect of childhood on self-perceptions around illness. Additionally, SBC represents a recurrent episodic illness and may not, therefore, be perceived as a chronic disease despite the long disease course.

Children and parents reported that receiving a SBC diagnosis did not change their level of activity or participation as an individual or a family and that they were eager to return to a previous level of sport following healing of pathological fractures. At the time of presentation there was an initial sharp reduction in activity and function but function returned as the fracture healed and activity participation then followed. The quantitative data demonstrated that children achieved a level of physical function participation similar to that of children with no medical conditions. Although children returned to activity, the qualitative data demonstrated that the way they thought about participation, especially in the period of time following fractures, was affected. Children readily acknowledged that they considered their SBC and the risk of re-injury or re-fracture, often immediately prior to participation, and that they exercised more caution while playing.

The consideration of patients' subjective evaluation of their disease and outcomes has become widespread in medicine and surgery in recent decades,<sup>24</sup> with a significant increase in the development and use of patient-reported outcome measures (PROMs).<sup>25</sup> PROMs and patient-centred outcomes provide the patient's perspective and are essential in supporting patient-centred care. While these tools, such as the ASK, take steps to provide a quantitative measure of some subjective domains, they also face limitations. Function is clearly an important outcome of SBC for both patients and physicians. However, the ASK was not able to capture the subtle changes in activity and thought about activity experienced by children during the course of their SBC as reported in the qualitative interviews. Instead, the results of the qualitative interviews revealed that the way they thought about participating in sport and play might be temporarily affected and resolved following cyst resolution.

The use of a mixed methods approach allowed us to more fully understand the impact of SBC on children and their families. The qualitative and the quantitative data each provided important information about the outcomes and effects of the disease. While these data sets could stand alone in the benefit they provide to understanding this disease, the integration of the two using mixed methods allowed each to bolster the other and shed further light in explaining the results leading to a more comprehensive understanding of the impact of SBC.

The main limitation of this study is the potential difference between the interview and trial cohorts. This was addressed by comparing ASK scores and demographic data, however, the interview cohort was relatively small, and some differences may not be captured. Additionally, a subgroup analysis comparing function related with upper *versus* lower extremity SBCs was not possible due to the limited numbers in this study. This study focused on functional limitations and the impact of SBCs and, therefore, other parameters such as a frequency of pathological fractures and number of interventions were beyond its scope. However, previous data has shown no association between pathological fractures subsequent to diagnosis and ASK scores.<sup>9</sup> This study raised many interesting areas for potential further research into the factors that may impact outcomes of SBC including further investigation into the effects of cyst location, number of fractures and number of interventions required for treatment.

One implication of this study is that function scores such as the ASK alone may not be appropriate for measuring the outcomes of SBC. Children in this study had high function with nearly 40% achieving a perfect ASK score. However, the ASK does not take into consideration anxiety, which was a common theme in children. One way of addressing the limitations of the ASK in cases of SBC may be combining it with an anxiety measure related to physical illness or physical function.

## Conclusion

Children and parents generally reported no change in level of activity participation due to SBC beyond the immediate phase of fracture recovery. This finding corresponds with children not viewing themselves as having a chronic illness with medical needs. In the future, multiple outcome tools with a broader range of domains, specifically including anxiety, may help to better assess outcomes of SBC, including illness perceptions and physical function.

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## COMPLIANCE WITH ETHICAL STANDARDS

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### ETHICAL STATEMENT

**Ethical approval:** This study received approval from our institution's Children Research Ethics Board and all procedures performed involving human participants were in accordance with the ethical standards of the 1964 Helsinki declaration and its later amendments.

**Informed consent:** Informed consent was received prior to participation.

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### AUTHOR CONTRIBUTIONS

AB: Study design, Performed measurements, Statistical analysis, Manuscript preparation.  
BF: Study design.

AD: Study design, Manuscript preparation.

CF: Study design, Qualitative analysis, Manuscript preparation.

JW: Study design, Manuscript preparation.

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