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REGULAR ARTICLE

Health-related quality of life in Swedish children and adolescents with limb reduction deficiency

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ABSTRACT

Aim: To investigate health-related quality of life (HRQoL) in young persons with limb reduction deficiency (LRD).

Methods: One hundred and forty children with LRD aged 8–16 answered the DISABKIDS questionnaire for children with chronic health conditions. Of their parents, 137 answered a corresponding questionnaire concerning their child.

Results: Compared to reference data from children with other health conditions, children with LRD showed higher overall HRQoL and higher HRQoL in all subscales except social exclusion. Overall, the results were not related to gender or age, but girls with longitudinal, bilateral or lower LRD reported significantly lower HRQoL in most subscales than girls with other forms of LRD. Unexpected attention and perceived physical appearance had a significant impact on HRQoL. There was poor agreement between parent and child report of the child's HRQoL.

Conclusion: Children and adolescents with limb reduction deficiency have a better HRQoL than children with other health conditions but there are subgroups of children who experience a significantly lower HRQoL than their peers. The difference between parent and child ratings should be considered in clinical practice.

The prevalence of limb reduction deficiency (LRD) in Sweden is 6.3 per 10 000 births (1) and results in 60–70 newborns yearly with LRD. The impact of LRD on everyday life varies, ranging from practical limitations in daily activities to negative effects on psychological and social well-being.

The role of LRD for well-being and mental health is only partly understood although it has been studied from different points of view. Perceived physical appearance (2–5), unexpected attention and practical problems (2,6,7) seem to have an impact on psychosocial well-being in children with LRD. Some previous studies show that children and adolescents with LRD have more behavioural and emotional problems and lower social competence than the normative community sample. As a consequence, Varni and Setoguchi consider children with LRD to be at risk for emotional distress and social adjustment problems and discuss the condition in terms of a 'new hidden morbidity' among these children (8). A contradicting study (7) shows that children with upper limb deficiency exhibit as good mental health as their able-bodied peers but girls and older children display a wider variety of problems than the group as a whole.

Most studies on health in children with LRD are proxy reports, obtaining information from parents only. Earlier studies have shown that parents of healthy children usually overestimate quality of life levels compared to that reported by their child, whereas parents of children with a health condition tend to underestimate the child's quality of life. Furthermore, the divergence seems to be larger in emotional rather than in physical domains (9–11). Consequently, it is important to study the child's own impression to understand the 'meaning of LRD'. In most of the previous studies the instruments used are symptom-oriented and designed for measuring psychiatric problems. However, these measures may not capture the whole range of ways in which a child may be affected. Studies of health-related quality of life (HRQoL) provide another way of conceptualizing the consequence of a health condition. This has been studied in

Abbreviations
HRQoL, health-related quality of life; LRD, limb reduction deficiency present at birth.

many children with health conditions but despite the earlier interest in health condition in children with LRD (12), HRQoL in this group has only been studied in children with upper LRD (13). The aims of this study were thus to investigate HRQoL in children with various forms of LRD, and to compare this to the HRQoL in children with other health conditions.

Further aims were as follows: a) to study the effect on HRQoL in children with LRD of the following factors: i) gender and age; ii) type, extent, site and level of the deficiency; and iii) perceived physical appearance and unexpected attention; and b) to study the consistency between child and parent report of the HRQoL of children with LRD.

PATIENTS AND METHODS

A cross-sectional multicentre design was chosen for this study. Participating units were as follows: (i) The Limb Deficiency and Arm Prostheses Centre, Örebro University Hospital, Örebro; (ii) The Arm Prostheses Centre, Sahlgrenska University Hospital, Göteborg; (iii) The BEDA-team, department of Orthopaedics, Lund University Hospital, Lund; (iv) Astrid Lindgrens Childrens Hospital, Karolinska Hospital, Stockholm; and, (v) Center for Arm Amputees, Red Cross Hospital, Stockholm.

Before initiation, the study was approved by the Regional Ethical Review Board in Uppsala, Sweden. Informed consent was obtained from all participants.

Exclusion criterion

To prevent possible confounding effects from other health conditions that may have an impact on quality of life, children with syndromes such as TAR (Thrombocytopenia and Absent Radius syndrome) were excluded. Also, children who were adopted from foreign countries and children and parents who were unable to read or understand the questions correctly because of poor language skills were excluded.

Participants

The subjects comprised 140 children and adolescents (boys n = 72, 51%) aged 8–16 (mean age: boys 11.7 years, girls 11.8 years) with LRD. The age limits, 8–16 years, were chosen to match the measurement requirements. The parents of 137 of the children also completed the questionnaire. Additionally, five parents completed the questionnaire without their child participating. In all, 142 parents participated in this study. The sample comprised all children eligible at the participating units during the study period (January 2007 to June 2008). Ninety-seven (69%) children had a transverse type and 43 (31%) had a longitudinal type of deficiency; 120 (86%) children had LRD in upper limbs, 11 (8%) in lower limbs and 9 (6%) in both upper and lower limbs. Most (121, 86%) of the children had unilateral deficiency (one affected limb), whereas 10 (7%) had bilateral deficiency (either both upper or both lower limbs affected) and 9 (6%) of the children had multiple deficiencies (at least one upper and one lower limb affected). The level of deficiency in children with unilateral upper limb malformation (n = 114) was above elbow 3 (3%), below elbow 66 (58%) and partial hand 45 (39%). The children with unilateral malformation in lower limb consisted of seven children, and they all had malformation below the knee.

Measurements

The Swedish version of DISABKIDS Chronic Generic Measure (DCGM-37) was selected for the study (14). This is a recently developed cross-cultural questionnaire for assessment of HRQoL in children and adolescents aged 8–16 with chronic health conditions, available in both self-completion and proxy versions (Appendix S1). Internal consistency values (Chronbachs' Alpha 0.70–0.87 in child version and 0.77–0.90 in proxy version) and test–retest reliability (0.71–0.83) has been established (14). The DISABKIDS is built of six subscales, *Physical limitation, Emotion, Independence, Social inclusion, Social exclusion* and *Treatment*, with six or seven statements in each. The statements are answered on a five-point Likert scale. Besides a score on each individual subscale, an overall HRQoL score can be computed from the answers. In this study the *Treatment* subscale was excluded because no child scored any item on this.

Reference data including European children with the following conditions, asthma, arthritis, atopic dermatitis, diabetes, cystic fibrosis, cerebral palsy and epilepsy, originating from the DISABKIDS manual (14), were used for comparison.

As reported earlier, perceived physical appearance (2–5) and unexpected attention (2,6,7) have an influence on psychosocial well-being in persons with LRD. To study the impact of these factors on HRQoL the following two study-specific questions were formulated:

'Do you think you are as good-looking as other children/youths?

'Does it bother you if other people ask you questions or stare at you because of your condition?'

To correspond with the DISABKIDS response alternatives, the same five response options were available for the study-specific questions. These were: 'Always', 'Very often', 'Quite often', 'Seldom' and 'Never'.

The children were divided into age groups corresponding to the DISABKIDS reference group, namely child (8–12 years) and adolescent (13–16 years) (14). Classification of type, location (extent and site) and level of deficiency were done according to ISO 8548-1:89 (15). Because of the small number of children in the other categories, analysis of the effect of level of deficiency on HRQoL was performed only in children with a unilateral upper LRD.

Procedure

To secure a similar method for data collection, comprehensive written information about the study protocol was shared between members of the participating units. One main issue was to ensure that the participants would answer the questionnaire independently of each other. Data were collected from January 2007 to June 2008 by

one of two procedures: (i) Consecutively, at the time of a clinic appointment, the child and parent received information and verbal consent was obtained from both of them; (ii) Twenty families (14.3%) had no scheduled appointment during the data-collecting period. They were contacted over the telephone by the researchers and consent was obtained from the parent and the child. The children who had no scheduled clinic appointment completed the questionnaire and the study-specific questions at the school nurse's clinic with the assistance of a teacher or a local occupational therapist. In accordance with the study protocol, all these cases followed the same written instructions. Parents of these children completed the questionnaire at home by themselves.

Statistical analyses

In accordance with the DISABKIDS manual (14), the data were expressed as indices derived from a summation and transformation (0–100) of the scores from each domain. Subsequently, these data were compared with the reference data. Student's one-sample *t*-tests with hypothesized mean values obtained from the reference data were performed to test for differences between data for the LRD sample and the reference data.

To detect any possible effects of gender, age, disability-specific characteristics, or perceived physical appearance and unexpected attention on HRQoL in the LRD sample, one-way analyses of variance (ANOVAs) were performed. Because of the small number of children with bilateral and lower LRD, ANOVAs to study the effect of level of deficiency were performed on children with unilateral upper LRD only (n = 114). p-values lower than 0.05 were accepted as statistically significant.

To test for differences between child and parent reports, Student's paired sample *t*-tests were performed. However, when the consistency between child and parent report is considered, hypothesis-testing is not of primary interest. Here, the main focus is on the agreement between the two reports. Hence, the Intraclass Correlation Coefficient (ICC) (16) was used as a measure of agreement. The ICC's were estimated from one-way ANOVA estimates and were supplemented with 95% confidence intervals. It usually ranges between 0 and 1.0. The guidelines for interpretation of agreement proposed by Fleiss (17) were used to interpret the strength of the agreement. In accordance with Fleiss, agreement below ICC 0.40 was considered poor, 0.40 – 0.75 represents fair to good agreement, and 0.75 and above represents excellent agreement. All calculations were performed using SPSS version 15 (SPSS Inc., Chicago, IL, USA).

RESULTS

When compared to the European reference population of children and adolescents with a chronic health condition, Swedish LRD children, adolescents and their parents, respectively, reported significantly higher ($p < 0.000$) overall HRQoL and specific HRQoL in all subscales except in *Social exclusion* ($p = 0.11$, $p = 0.94$) (Table 1).

Table 1 Comparison of self-reported and parent-reported HRQoL between Swedish childrenwith limb reduction deficiency and European children with common chronic health conditions

	Children's report of their own HRQoL		Parents' report of HRQoL of their child	
	Swedish	European†	Swedish	European†
	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)
	n = 140	n = 1152	n = 142	n = 1152
Overall HRQoL	82 (12)	77* (14)	80 (12)	75* (15)
Physical limitation	82 (13)	74* (18)	80 (14)	70* (18)
Emotions	85 (16)	77* (21)	80 (17)	72* (20)
Independence	82 (13)	77* (18)	81 (12)	77* (17)
Social inclusion	80 (14)	75* (18)	80 (15)	74* (18)
Social exclusion	83 (17)	85 (16)	81 (15)	81 (17)

Note: Possible range 0–100. Higher score indicates better HRQoL.
*$p < 0.05$ (One-sample *t*-test).
†Common chronic conditions: asthma, arthritis, atopic dermatitis, diabetes, cystic fibrosis, cerebral palsy, epilepsy.
HRQoL = health-related quality of life.

Effect of gender, age, type, extent, site and level of deficiency

When age or gender was used as the independent variable in the ANOVAs, there was no statistically significant effect on HRQoL. Furthermore, in the group as a whole, there were no effects of type, extent or site of deficiency on HRQoL. Nor was there any significant effect on HRQoL in children with unilateral upper LRD based on level of deficiency. When analysing subgroups by gender, no significant effects could be seen in boys. However, girls with longitudinal LRD had significantly ($p = 0.03$) lower scores in the *Emotion* subscale than girls with transverse LRD (Table S1). Likewise, as seen in Table S1, when using extent of deficiency as the independent variable in the ANOVAs, girls with bilateral LRD had significantly lower overall HRQoL ($p = 0.01$), *Physical limitations* ($p < 0.01$) and *Social inclusion* ($p < 0.01$) than girls with unilateral or multiple LRD. There was a similar effect of site of deficiency showing that girls with LRD in lower limbs have significantly lower overall HRQoL ($p = 0.01$) and lower scores in *Physical limitations* ($p < 0.01$), *Emotions* ($p = 0.04$) and *Social inclusion* ($p < 0.01$) than girls with LRD in upper limbs and girls with multiple LRD (Table S1).

Effect of Perceived physical appearance and unexpected attention

Boys and girls who considered themselves as good-looking as others reported higher HRQoL than those who considered themselves less attractive. When degree of perceived physical appearance was used as the independent variable in the ANOVAs, there were significantly lower scores in the overall score ($p < 0.01$) and all subscales ($p < 0.01$ – $p = 0.001$) of DISABKIDS in boys who 'never' or 'seldom' perceive themselves as good-looking as others, than boys who 'always' consider themselves as good-looking as others. A similar effect was found in girls who 'never' think they are

as good-looking as others, with significantly lower HRQoL in the *Emotion* subscale ($p = 0.01$) than girls who 'always' consider themselves as good-looking as others.

Both boys and girls who experience unexpected attention negatively reported lower HRQoL than those who were not bothered by the attention. Those boys and girls who are 'always' or 'very often' bothered by unexpected attention have significantly lower overall HRQoL ($p < 0.01$, $p < 0.01$) and lower scores in *Physical limitations* ($p < 0.01$, $p < 0.01$), *Emotions* ($p = 0.02$, $p < 0.01$), and *Social exclusion* ($p < 0.01$, $p < 0.01$), respectively. These boys also score significantly lower in *Social inclusion* ($p < 0.01$), and girls score significantly lower in *Independence* ($p < 0.01$) than those children who are 'never' or 'seldom' bothered by unexpected attention.

Consistency between child and parent report

As expected, the agreement between parent and child reports of HRQoL was mostly poor (Table S2). Parents scored their child's overall HRQoL significantly lower ($p = 0.02$) and gave lower scores in *Physical limitation* ($p = 0.01$) and *Emotions* ($p < 0.01$) modules for girls, and in the *Emotions* module ($p = 0.02$) for younger children. They gave lower scores in all submodules for older children ($p < 0.01 - 0.02$), giving a significantly lower overall HRQoL ($p < 0.01$).

DISCUSSION

The main finding in this study is that children with LRD report significantly higher HRQoL than children with other chronic health conditions. This confirms earlier findings of psychosocial adjustment and HRQoL in children with upper LRD (7,13), where they exhibit similar health to their nondisabled peers. Together, these findings may support the conclusion that, in general, the condition of LRD does not have a large impact on HRQoL.

However, the results on the *Social exclusion* subscale, in which the children with LRD had scores similar to the reference group (Table 1), indicate a social impact of the deficiency. This subscale consists of two concepts, *stigma* and *feeling left out* (14). The lives of persons with a disability are often connected to different valuations attributed to body forms (18). Hence, persons with a disability may feel stigmatized, or left out. This may be analogous to previous studies where increased withdrawal is reported among children of both genders with LRD (7). The social impact of LRD is also demonstrated by the answers to the study-specific questions. These results confirm previous research (6) indicating that unexpected attention and perceived physical appearance are associated with self-esteem in children with LRD. Furthermore, based on the present results, it seems as if these aspects of the deficiency have an impact on both boys and girls. Hence, unexpected attention and perceived physical appearance seem to be more strongly associated with HRQoL than localization, laterality or type of deficiency, which are only associated with HRQoL for girls. This relation between physical appearance and HRQoL has been demonstrated in children with other health conditions that have an impact on the appearance, e.g. haemangioma or oral clefts (19,20).

Analysis of subgroups revealed that girls with longitudinal, bilateral or lower LRD display lower HRQoL scores than girls with other forms of LRD in both overall HRQoL and in the *Physical limitation*, *Emotion* and *Social inclusion* subscales (Table S1). This contradicts Varni's results (8), which suggested that the degree of limb loss has no impact on psychosocial adjustment in children with amputation or limb deficiency present at birth. This needs to be studied further.

Disagreement between generations was expected (9–11). This is indeed shown in the results, where agreement between children and parents is poor (Table S2). The results from this study are similar to the results from Sheffler et al. (11), showing an inconsistency especially in girls and adolescents. It seems that parents tend to overemphasize problems and obstacles caused by the deficiency compared to their child's own ratings. The difference between parent and child ratings demonstrates the importance of studying the children's own expression of their perceived quality of life and should be taken into consideration in clinical practice. However, despite the differences we found between parents and children's reports of HRQoL in LRD (Table S2), parents' ratings are consistently higher than the reference population, supporting the difference between the groups of children (Table 1).

In earlier studies on children with various forms of LRD, symptom-oriented methods were used (7,8). To study the impact of LRD from a somewhat different angle, similar to James et al. (13) we chose to measure the HRQoL. However, in contrast to the previous study, we chose a measure developed in Europe, the DISABKIDS questionnaire (14). A recent study on HRQoL in Swedish children with diabetes (21) confirms the applicability of the DISABKIDS to a Swedish population. In their study, Chaplin et al. found gender and age differences in HRQoL. Another populations-based study carried out on a cross-section of children and adolescents in Europe (22) showed a decrease in HRQoL during adolescence. Other studies (7,23) in children with LRD have reported more psychosocial adjustment problems among girls and adolescents. Hence, we expected the HRQoL in children with LRD to be related to age and gender, but the results from this study show no difference between these groups. The reason for this may be that the method itself, DISABKIDS, does not capture the age or gender differences in this specific population. As demonstrated in the results (Table 1), children with LRD seem to have a high HRQoL. It is plausible that, at a high level of HRQoL, DISABKIDS cannot capture the age or gender differences because of ceiling effects. Further studies are needed to confirm this.

Beside the diagnosis-related difference, there are other possible explanations for the high level of HRQoL in this study. One reason could be the characteristics of the sample. Despite the fact that the number of participants in this study is relatively large, the participants only represent

approximately 25% of the Swedish age-group population with LRD (24). Furthermore, the representation of different types of LRD is skewed. Upper LRD constitutes approximately two-thirds of LRD cases in the population (25,26) whereas in this study the proportion was greater (86%). This could have had an effect on the results. Another issue that arises when analysing the results is that, as the 140 children who participated in this study were all patients at one of the participating units, they were receiving highly specialized team support. Both the high HRQoL in the study group and the unexpected absence of age or gender differences in this study may be attributed to the high quality of treatment and rehabilitation. A comparative study comprising a representative sample of children with LRD who are patients at the specialized centres to an equal sample of children with LRD who are nonpatients would highlight both these issues.

In conclusion, children and adolescents with LRD have a higher overall HRQoL than children of the same ages with other chronic conditions. There are, however, subgroups of children with LRD who experience a significantly lower health-related quality of life than their peers with LRD. They need to be evaluated and treated more carefully. The difference between parent and child ratings should be considered in clinical practice.

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SUPPORTING INFORMATION

Additional Supporting Information may be found in the online version of this article:

Appendix S1 DISABKIDS subscales and exemplification for each of them.

Table S1 Means and SD (within parentheses) of self-reported HRQL in girls aged 8–16 years with limb reduction deficiency, broken down by deficiency category.

Table S2 Intra-class correlation coefficient with 95% CI (within parentheses) of agreement between self-reported and parent-reported HRQoL.

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