

ORIGINAL REPORT

## PARTICIPATION AND HEALTH-RELATED QUALITY OF LIFE OF DUTCH CHILDREN AND ADOLESCENTS WITH CONGENITAL LOWER LIMB DEFICIENCIES

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**Objectives:** To describe participation and health-related quality of life of Dutch children and adolescents with congenital lower limb deficiencies in comparison with typically developing children, and to explore differences between various degrees of limb loss and between parental reports and self-reports on health-related quality of life.

**Design:** Cross-sectional study.

**Methods:** Participation was assessed with the Children's Assessment of Participation and Enjoyment questionnaire, and health-related quality of life with the KIDSCREEN-52 questionnaire, both as parental reports and self-reports, for 56 children and adolescents with congenital lower limb deficiencies, aged 8–18 years.

**Results:** Participation and health-related quality of life of children and adolescents with lower limb deficiencies (age range 8–18 years) did not differ from those of the reference group, except that the adolescents with lower limb deficiencies (age range 12–18 years) reported significantly ( $p < 0.05$ ) less diversity and lower intensity of social and skill-based activities. Degree of limb loss did not affect participation or health-related quality of life. Differences ( $p < 0.05$ ) between parental reports and self-reports for health-related quality of life were found for the "physical well-being", "moods and emotions" and "self-perception" domains. While parental reports were comparable to the adolescents' self-ratings, parents reported lower health-related quality of life in the "moods and emotions", "self-perception" and "autonomy" domains for their younger children.

**Conclusion:** While the participation and perceived health-related quality of life of Dutch children with lower limb deficiencies do not differ from those among typically developing children, the participation of adolescents with lower limb deficiencies is characterized by less diversity, with less interaction in social and skill-based activities.

**Key words:** adolescents; children; lower limb deficiencies; participation; health-related quality of life.

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### INTRODUCTION

Congenital lower limb deficiency (LLD) is a chronic disorder occurring in 2 out of 10,000 live births in the Netherlands. Forty percent of children with congenital LLD have both legs affected; 30% also have defects of one or both upper limbs (1). Although it appears that children with LLD live lives comparable to those of their peers, they are limited in terms of physical activities by having to depend on prosthetic devices (2, 3). Apart from physical limitations, children with LLD also differ from typically developing children in outward appearance. Since they are physically and visibly different, it has been suggested that children and adolescents with congenital LLD are at risk for psychosocial maladjustment (4, 5).

Participation and health-related quality of life (HRQoL) are two key concepts in paediatric research that improve our understanding of what parents, professionals and policymakers try to achieve for children with disabilities (6). Although both concepts are considered essential outcomes in describing health status and assessing interventions, the literature provides limited data on how children with congenital limb deficiencies participate and how they perceive their HRQoL (7).

In the framework of the International Classification of Functioning, Disability and Health (ICF), participation is one of the components describing function and health status, and is influenced by environmental and personal factors (8). Participation is defined by the World Health Organization (WHO) as involvement in life situations or being involved in everyday activities (8). Children's participation can be influenced by perceptions of self-competence and the presence of supportive environments (9–14). In general, participation in activities increases during childhood as the children mature and their skills develop (15, 16). However, it has also been found that, as children grow into adolescence, the number of activities they participate in decreases (17).

Another concept, pertaining to all domains of the ICF framework, is quality of life (QoL) or HRQoL (18). HRQoL refers to the assessment of various aspects of health from the patient's point of view and includes physical, mental and social well-being and functioning (19). Self-reported measures of HRQoL have been recommended even for children, when possible (20). Evidence has shown that children can reliably report on their

HRQoL, provided that their emotional development, cognitive ability and reading level are taken into account (21). Assessing children's HRQoL using both parental reports and self-reports is thought to offer valuable complementary information, as it can reflect meaningful differences (19). Parents of healthy children tend to report higher HRQoL than the children themselves, whereas parents of children with chronic conditions have a tendency to rate their children's HRQoL below the children's own rating (22). No consistent findings have been reported on factors influencing the agreement between child and parent (23), although poor parental well-being and children being in pain are known to have a negative effect on parents' perceptions of their children's QoL (23).

To date, no studies are available on the way that children and adolescents with LLD participate and how they perceive their HRQoL. The objective of the present study was to describe the self-reported participation and HRQoL of children and adolescents with congenital LLD in comparison with those of typically developing children. Differences between various degrees of limb loss were explored, as well as differences between parental reports and self-reports on the child's HRQoL.

METHODS

This cross-sectional study of children and adolescents with LLD was based on questionnaires completed as self-reports on participation and HRQoL and proxy reports by the parents on their children's HRQoL.

Ethical approval was received from the medical ethics committee at De Hoogstraat Rehabilitation Center. All parents and children gave written informed consent before participating in the study.

Participants

Children and adolescents with LLD, aged between 8 and 18 years, were recruited from the Rehabilitation Center De Hoogstraat in Utrecht, The Netherlands. The inclusion criterion was congenital LLD, with or without arm deficiencies. Children with cognitive impairment or syndromal disorders were excluded from the study.

Children/adolescents and their parents were contacted by sending them a letter offering information about the study. Those who did not wish to participate in the study were asked to return a reply card. After both the children and the parents who wanted to participate had signed and returned an informed consent form, the questionnaires were sent to the participants by post. Parents were allowed to assist their children in completing the questionnaires. Information about the degree of limb loss was obtained from the medical records of De Hoogstraat Rehabilitation Centre. For the purpose of this study, they were recorded as unilateral LLD, bilateral LLD and LLD in combination with arm deficiency.

Measures

Participation was measured using the Children's Assessment of Participation and Enjoyment (CAPE) (24). The CAPE is a self-report measure of children's participation in recreation and leisure activities outside mandated school activities. It can be used for children aged 6–18 years with or without disabilities (24). It takes approximately 30–45 min to complete. The CAPE contains 55 items, which are presented as drawings and texts providing information on 3 aspects of participation: diversity, intensity and enjoyment. Diversity scores indicate the number of activities (maximum 55) performed by the child over the past 4 months. Intensity scores indicate the frequency with which the child participates in activities. More diverse and frequent activity participation is reflected by higher diversity and

intensity scores. The CAPE includes 5 activity types: recreational, active physical, social, skill-based and self-improvement activities. For the purpose of the present study, overall diversity and intensity scores for the 5 types of activities were used to describe the level of participation of children and adolescents with LLD. Psychometric evaluation of the CAPE has demonstrated satisfactory validity and reliability (24). The present study used the Dutch version, which has comparable psychometric properties (25). Comparative data on the participation of typically developing children were obtained from a convenience sample of 158 Dutch children, aged 6 to 18 years (mean age 12 years; standard deviation (SD) 3), recruited from 4 mainstream schools in the Netherlands (25).

Quality of life was measured using the Dutch version of KID-SCREEN, a 52-item generic HRQoL measure (26). This questionnaire was designed for reporting by children and parents and can be used for healthy and chronically ill children and adolescents aged 8–18 years (26). The time required for administration is 15–20 min. Ten domains of HRQoL are assessed: physical well-being, psychological well-being, moods & emotions, self-perception, autonomy, parental relations & home life, financial resources, peers & social support, school environment, and bullying. The questionnaire uses 5-point Likert scales with two different sets of responses: never, seldom, quite often, very often, always and not at all, slightly, moderately, very, and extremely. For each domain, the relevant items are summed and scaled to yield a score with a range of 0–100, with higher scores indicating better QoL. No overall score can be calculated. The KIDSCREEN is a well-validated measurement tool that allows comparisons with the Dutch general population (27). Reference data are available for gender and two age groups: 8–11 years and 12–18 years (28).

Statistical analysis

Descriptive statistics (frequency, means, SD) were used to describe the sample and the children's diversity and intensity of participation. Data for children and adolescents with LLD were compared with reference data using Student *t*-tests. Different degrees of limb loss (unilateral, bilateral and combinations of limb deficiencies) were compared using Mann–Whitney *U* tests because of the small sample sizes.

HRQoL scores for each domain for both parents and children are presented as mean and SD. We used the recommended thresholds, which are defined as the mean (50) ± half of the standard deviation (28). Comparison with reference data was done by a 1-sample *t*-test using the *t*-values of the reference group as the test value. The scores of the children and their parents were compared using a paired *t*-test to determine if there were statistically significant differences between these groups. All analyses were performed with SPSS version 15.

Table I. Demographic characteristics of participants with limb deficiencies (n = 56)

Demographic characteristics	
Age, years, mean (SD)	12.6 (3.1)
8–11 years, n (%)	23 (41)
12–18 years, n (%)	33 (59)
Gender, n (%)	
Boys	34 (61)
Girls	22 (39)
Degree of lower limb loss, n (%)	
Unilateral	34 (61)
Bilateral	9 (16)
Combined with upper limb loss	13 (23)
Type of school, n (%)	
Mainstream	51 (91)
Special	5 (9)
Parent proxy responders, n (%)	
Mother	49 (88)
Father	7 (12)

SD: standard deviation.

Table II. Diversity and intensity of Children's Assessment of Participation and Enjoyment (CAPE) activities in children and adolescents with lower limb deficiencies (LLD) compared with the reference group

	LLD 8-18 years (n=53)	Reference group 8-18 years (n=158)	LLD 8-11 years (n=21)	Reference group 8-11 years (n=92)	LLD 12-18 years (n=32)	Reference group 12-18 years (n=66)
CAPE activities	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)
Diversity (0-55)	25.8 (6.4)	27.1 (5.8)	28.1 (6.8)	26.2 (6.0)	24.2* (5.8)	28.6 (5.2)
Intensity (1-7):						
Recreational	3.2 (1.2)	3.4 (1.1)	4.1 (1.0)	3.9 (1.0)	2.7 (1.0)	2.6 (0.9)
Physical	1.5 (0.6)	1.6 (0.8)	1.5 (0.6)	1.5 (0.7)	1.5 (0.6)	1.8 (0.7)
Social	3.0 (0.9)	2.9 (1.0)	2.8 (0.9)	2.4 (0.9)	3.2* (0.8)	3.5 (0.7)
Skill-based	1.0 (0.9)	1.1 (0.8)	1.3 (1.0)	1.0 (0.8)	0.8* (0.8)	1.3 (0.8)
Self-improvement	2.2 (0.9)	2.4 (1.1)	1.9 (0.7)	2.1 (1.1)	2.3 (0.9)	2.7 (1.0)

\*p<0.05, compared with reference group. SD: standard deviation.

RESULTS

Sixty-four children and their parents were invited to take part in the study. One family declined to participate by returning the reply card, while 7 families declined after being contacted by telephone. The questionnaires returned by 2 families were lost in the post. Both families agreed to complete KIDSCREEN-52 a second time, but not the CAPE, as this was too time-consuming. One family refused to complete the CAPE because they felt that the questionnaire did not fit their (18-year-old) daughter's lifestyle. As a result, KIDSCREEN-52 questionnaires were completed by 56 parents and their children with LLD (88%), while the CAPE was completed by 53 parents and their children (83%). Nineteen (36%) children required help in completing the CAPE, 15 of whom were younger than 12 years. In all these cases, assistance was provided by the mothers. No children required help in completing the KIDSCREEN-52 questionnaires.

The demographic characteristics of the participants with LLD are summarized in Table I.

Participation

Children and adolescents with LLD (8-18 years) participated in many different activities (Table II). No differences in di-

versity of activities were reported for children with LLD (age range 8-11 years) in comparison with the reference group. Adolescents (age range 12-18 years) with LLD engaged in statistically significantly less diverse activities than the reference group (p=0.001).

Intensity of participation as reported by all children and adolescents with LLD (age range 8-18 years) did not differ significantly from that in the reference group. Children with LLD (age range 8-11 years) reported no differences in intensity of participation compared with the reference data. However, adolescents with LLD had statistically significantly lower intensity of social activities (p=0.041) and skill-based activities (p=0.008) compared with the reference group of the same age.

No differences were found between children and adolescents with different degrees of limb loss, in terms of either diversity or intensity of participation.

Health-related quality of life

Perceptions of children and adolescents with LLD (age range 8-18 years) of their HRQoL were comparable to those for the Dutch reference group (Table III). Children (age range 8-11 years) and adolescents (age range 12-18 years) with LLD re-

Table III. Health-related quality of life dimensions based on parental report and self-report by children and adolescents with lower limb deficiencies (LLD), compared with the reference group

	LLD 8-18 years (n=56)	Reference group 8-18 years	LLD 8-18 years (n=56)	LLD 8-11 years (n=23)	Reference group 8-11 years	LLD 8-11 years (n=23)	LLD 12-18 years (n=33)	Reference group 12-18 years	LLD 12-18 years (n=33)
KIDSCREEN-52 dimensions	Parental Self-report Mean (SD)	Parental Self-report Mean (SD)	Parental report Mean (SD)	Self-report Mean (SD)	Self-report Mean (SD)	Parental report Mean (SD)	Self-report Mean (SD)	Self-report Mean (SD)	Parental report Mean (SD)
Physical Well-being	52.4 (10.8)*	52.9 (10.0)	49.7 (10.2)	56.6 (10.9)	57.3 (9.5)	52.1 (11.2)	49.5 (9.8)	50.4 (9.4)	48.0 (9.3)
Psychological Well-being	53.4 (8.9)	53.3 (8.9)	53.0 (8.9)	55.4 (7.8)	55.8 (9.2)	54.9 (8.9)	52.0 (9.5)	51.6 (8.5)	51.7 (8.8)
Moods & Emotions	52.4 (10.1)*	51.4 (9.9)	47.7 (10.8)	53.4 (10.9)*	52.6 (9.7)	46.6 (10.8)	51.7 (9.6)	50.7 (9.9)	48.4 (10.8)
Self-Perception	51.7 (8.4)*	52.2 (9.9)	47.4 (7.3)	53.9 (8.5)*	56.9 (9.6)	47.3 (6.8)	50.1 (8.1)	49.6 (9.0)	47.5 (7.7)
Autonomy	53.1 (7.6)	54.4 (8.9)	51.0 (7.3)	54.1 (7.7)*	56.2 (8.5)	51.0 (6.4)	52.4 (7.5)	53.3 (9.0)	50.9 (7.9)
Parent Relation & Home Life	52.8 (8.5)	53.2 (9.0)	52.9 (8.6)	52.7 (8.2)	55.2 (8.3)	53.4 (9.3)	52.8 (8.9)	52.0 (9.2)	52.6 (8.1)
Social Support & Peers	51.5 (12.1)	52.4 (9.3)	50.4 (10.8)	51.0 (13.5)	53.1 (9.3)	49.5 (12.9)	51.8 (11.2)	52.0 (9.3)	50.9 (9.2)
School Environment	52.7 (9.7)	53.2 (9.8)	51.9 (8.9)	56.7 (8.9)	58.1 (10.1)	53.8 (8.3)	49.9 (9.3)	50.4 (8.4)	50.6 (9.2)
Social Acceptance & Bullying	50.4 (9.9)	48.4 (10.2)	48.6 (11.3)	48.7 (10.8)	47.8 (10.8)	45.1 (12.6)	50.3 (8.8)	48.7 (9.8)	51.1 (9.7)
Financial Resources	54.6 (8.7)	52.1 (9.5)	55.4 (8.6)	52.2 (10.3)	51.3 (10.4)	53.6 (9.4)	54.8 (11.3)	52.5 (9.0)	52.6 (8.1)

\*p<0.05, compared with parental report. SD: standard deviation.

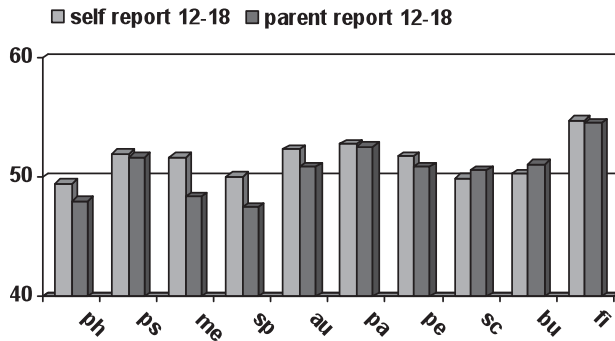


Fig. 1. KIDSCREEN-52 health-related quality of life scores by dimension for adolescents with lower limb deficiencies and their parents. Scales for each dimension: mean=50 in the reference population. ph: physical well-being; ps: psychological well-being; me: moods & emotions; sp: self-perception; au: autonomy; pa: parent relation & home life; pe: social support & peers; sc: school environment; bu: bullying; fi: financial resources.

ported HRQoL similar to that reported in the reference group. Subgroups with different degrees of limb loss did not differ from each other, and all were comparable to the reference group.

A comparison between parental reports and self-reports of HRQoL for all children and adolescents with LLD (age range 8–18 years) yielded statistically significant differences for the domains of “physical well-being” ( $p=0.045$ ), “moods and emotions” ( $p=0.006$ ) and “self-perception” ( $p=0.001$ ). The comparison of parents’ ratings with those of the younger children (age range 8–11 years) yielded statistically significant differences for the domains of “moods and emotions” ( $p=0.022$ ), “self-perception” ( $p=0.009$ ) and “autonomy” ( $p=0.044$ ). No statistically significant differences were found between parental reports and self-reports by the adolescents with LLD (Fig. 1).

## DISCUSSION

The objective of this study was to describe the participation and HRQoL of Dutch children and adolescents with congenital LLD in comparison with a reference group, using self-reports. Children’s and adolescents self-reports for HRQoL were compared with their parents’ proxy report. Key findings indicate that the overall participation and perceived HRQoL of Dutch children with LLD (age range 8–18 years) do not differ from those of the reference group. The participation of adolescents with LLD (age range 12–18 years) was found to be less diverse and less frequent as regards social and skill-based activities, while their HRQoL was comparable to that of typically developing children. Differences in degree of limb loss did not affect participation or HRQoL. Although the parents rated the HRQoL of their younger children (age range 8–11 years) lower than the children themselves, their HRQoL ratings for the adolescents (age range 12–18 years) were similar to the self-reports by the adolescents.

It is important to recognize that, despite their physical impairments, children with LLD, whether unilateral, bilateral

or in combination with arm deficiencies, participate fully in diverse activities and report comparable HRQoL as typically developing children. Children with LLD generally have a high level of physical activity and participate in a variety of sports (2, 3). It is known that higher levels of participation are associated with higher levels of physical, cognitive and communicative functioning (12). Our study excluded children with cognitive impairment and/or syndromal disorders. Ninety-one percent of our study population attended a mainstream school, indicating appropriate cognitive and communication skills.

Another finding from various studies is that children’s level of participation decreases with age (29) and that frequency of physical activity engagement decreases as adolescents grow older (30). The reduced participation we found among adolescents with LLD might be a result of growing up with a congenital physical impairment. This may have forced them to make choices regarding the number and type of activities earlier in their lives than adolescents in general do. They have grown accustomed to their abilities and disabilities and participate at a level that fits their lifestyle, which may explain the discrepancy between reduced participation and high reported HRQoL. The impact of their participation on the perceived HRQoL may have much to do with whether and how the children, despite their physical impairment, participate in activities that matter most to them (31).

Varni & Setoguchi (32) reported that adolescents with limb deficiencies tend to be more vulnerable to negative social perceptions and behaviours. Research among children with cerebral palsy found that negative reactions by others, such as bullying and staring, negatively influence their participation (33). Negative attitudes by others may also explain why adolescents with LLD participate in fewer diverse and social activities. More research is needed to explore factors influencing participation of adolescents with LLD.

Like other children with physical disabilities (34–36), children and adolescents with LLD perceive an HRQoL that is comparable to that of typically developing children. Parents, however, rated their child’s (age range 8–11 years) self-perception, autonomy and school environment lower than the child’s own rating. This suggests that parents have a more negative perception of how the limb deficiencies affect their children. This difference between parental and self-reports has also been found in other studies, in which parents of children with chronic conditions reported lower HRQoL than the children themselves (22). It has been suggested that children (age range 8–11 years) tend to have different response styles from those of their parents (37). On the other hand, HRQoL ratings by parents in our study were similar to those in the adolescents’ self-reports. This might imply that parents are more concerned about their younger children and that they develop more confidence in the well-being of their children as they grow older. This is an important finding for clinical practice. Parents can be reassured about the well-being of children with limb loss, and although the children need to learn to live with the limb deficiency, it appears that, overall, they do well.

Unlike our study, a study comparing children’s and parents’ reports on the QoL of children with below-the-elbow deficiency



found significant differences between the reports by parents and adolescents (38). The contrasting findings reflect the need to explore the effects of variables such as age, gender, child characteristics and parent-child relationships on the level of agreement between parents' and children's HRQoL reports.

Some comments can be made on the outcome measures we used, CAPE and KIDSCREEN-52. It is doubtful whether the CAPE is appropriate for the broad age span of 6–18 years. Adolescents do not always feel that they can relate to the activities described in the CAPE. Measurement of participation in at least two age bands seems more appropriate, given the growing autonomy experienced by adolescents (39). Although the CAPE objectively measures the level of participation, it does not show whether this level is perceived as good enough or as a limitation, from the perspective of the respondent. Various authors (6, 9) have argued that the subjective experience of participation is covered partly by the construct of HRQoL, which supports the use of KIDSCREEN-52 in the present study. In the study by Young et al. (40), the dimensions of KIDSCREEN-52 were compared with accounts of well-being by children with disabilities. The findings suggested that KIDSCREEN-52 were in good agreement with the children's accounts of their lives, though some specific aspects of life were missed, including home life, neighbourhood, siblings, pain and discomfort, inclusion and fairness in relationships, particularly peer relationships (40). It remains unclear whether all aspects of life that are important to children with LLD are represented by KIDSCREEN-52.

The strength of our study is that it is the first to explore the participation and HRQoL of children and adolescents with congenital LLD using validated measurement tools. Data on participation were collected by means of self-reports, and HRQoL was measured using both self-reports and parental reports. Both reports can be considered as complementary, providing a broad perspective on the HRQoL of children with LLD. All participants were recruited from a single-centre convenience sample, resulting in a high response rate. A broad geographic range was represented, as the Rehabilitation Center De Hoogstraat offers supra-regional rehabilitation services.

This study was subject to a few limitations. The first was the small sample sizes in the subgroups with different degrees of limb loss. In our study population, degree of limb loss did not affect participation or HRQoL. Although data were explored both in subgroups and at the individual level, no consistent differences were found in relation to the degree of limb loss. Although a lower HRQoL might be expected in the children with multiple limb deficiencies, the lack of statistical significance in our study may have been caused by the small number of children with multiple deficiencies. The second limitation was that we did not explore factors influencing participation and HRQoL. Although the study population was doing well on both measures, more research is needed to identify factors influencing participation and HRQoL in children and adolescents with LLD. It would also be interesting to know whether the children with LLD participate in the activities that they prefer or in activities that they are able to do. We believe that the statistically significantly lower participation scores of the

adolescents warrant the evaluation of individual scores, in order to determine whether they represent a problem for individuals. This would reflect the clinical relevance of the scores. We therefore recommend routine assessment of both outcomes, including children's own perception of their participation, to optimize service planning and development of intervention programmes for those families and children with LLD who do not participate to the extent that they would like to.

In conclusion, while the participation and perceived HRQoL of Dutch children with LLD (age range 8–18 years) are comparable to those of typically developing children, adolescents with LLD (age range 12–18 years) participate in less diverse activities with less interaction in social and skill-based activities. Parents tend to rate the HRQoL of their children aged 8–11 years lower than the children themselves. Degree of limb loss does not affect participation or the perception of the different dimensions of HRQoL.

These findings provide a better understanding of the participation and HRQoL of Dutch children and adolescents with congenital LLD. They can be used to reassure families and service providers that children and adolescents with congenital LLD have a full potential to participate in society and that they generally perceive their lives as satisfactory.

Routine assessment of both concepts is recommended in order to achieve optimal participation and HRQoL in children and adolescents with congenital LLD, providing a basis on which healthcare professionals, children and their families can plan their goals.

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