ORIGINAL ARTICLE

Poor Agreement on Health-Related Quality of Life Between Children With Congenital Hand Differences and Their Parents

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ABSTRACT. Ardon MS, Selles RW, Roebroeck ME, Hovius SE, Stam HJ, Janssen WG. Poor agreement on health-related quality of life between children with congenital hand differences and their parents. Arch Phys Med Rehabil 2012;93:641-6.

Objectives: To determine agreement between children with congenital hand differences (CHDs) and their parents on health-related quality of life (HRQOL) and to explore whether characteristic variables were associated with this agreement on different domains of HRQOL.

Design: Survey.

Setting: University hospital, outpatient clinic.

Participants: Children with CHD (age range, 10–14y; N=106).

Interventions: Not applicable.

Main Outcome Measure: Agreement on HRQOL was determined by comparing child self-reports and parent proxy-reports of the Pediatric Quality of Life Inventory 4.0 generic core scales, in Dutch. Agreement was examined both at group level and individual level.

Results: On a group level, children scored the same as their parents on a scale of 0 to 100 (physical health, 89.1±14.1 vs 88.0±15.6; psychosocial health, 80.6±13.4 vs 79.0±14.5; and total HRQOL, 83.5±12.3 vs 82.0±13.6). On an individual level, however, scoring was subject to high variation, with children reporting both higher and lower scores than their parent proxy. There were no major determinants for agreement; we only found that agreement was higher on emotional functioning in children with more affected fingers and on social functioning in bilaterally involved children.

Conclusions: In terms of mean group scores, 10- to 14-year-old children with CHD agree with their parents or proxy on the child's HRQOL. However, on an individual level, they disagree; on some subdomains limits of agreement are as large as 30 points on the 0 to 100 scale. Therefore, care should be taken in cases where children are unable to complete the questionnaire in choosing the parents' score as a representative substitute for the child's score.

Key Words: Child; Female; Hand deformities, congenital; Male; Quality of life; Questionnaires; Rehabilitation.

CONGENITAL HAND DIFFERENCES (CHDs) are not very common. Their prevalence is estimated at 16 per 10,000 live births, but varies within different populations and ethnic groups.[1] In frequency they are second to congenital heart malformations.[1]

The impact of health on a child's well-being is described by the World Health Organization (WHO) as health-related quality of life (HRQOL). HRQOL is defined as the individuals' perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, and concerns.[2] HRQOL has developed as an important outcome measure in pediatrics. In a previous study, our group reported on HRQOL in children with a CHD that is comparable with that of their healthy peers.[3] The study also found that HRQOL decreased in the presence of comorbidity but increased with higher ease of activity performance and that the severity, age, ethnicity, and surgery were associated factors.[3]

The WHO and the International Association for Child Psychology and Child Psychiatry recommended that children's quality of life measurements should be self-reported wherever possible.[2] In line with this and due to development of age-specific tools for children, HRQOL is increasingly measured from the child's point of view. A parallel version of the child's questionnaire for their parents allows comparison of both scores or can be used as an alternative if a child is unable or unwilling to score the questionnaires.[4-6] It is important to determine whether the proxy-report can be used interchangeably with the child's self-report and therefore can be used to assess the child's HRQOL when their self-report data cannot be obtained. In addition, while it has been shown that child characteristics such as age, sex, and severity of disease influence child-parent agreement,[7] to our knowledge, variables that influence agreement are not extensively studied in children with different kinds of CHD. Ylimaïnen,[5] Sheffler,[6] and colleagues found that children with below-the-elbow deficiency report better quality of life than their parents perceive. Sheffler[6] also found that factors influencing parent-child agreement on quality of life include age and use of a prosthesis.

The primary objectives of this cross-sectional study were to determine agreement between children 10 to 14 years old with a CHD and their parents and whether characteristic variables

List of Abbreviations

CHD	congenital hand difference
HRQOL	health-related quality of life
ICC	intraclass correlation coefficient
PedsQL	Pediatric Quality of Life Inventory
WHO	World Health Organization

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were associated with this agreement on different domains of HRQOL. We hypothesized that children and their parents would agree on all dimensions.

METHODS

This study used data from a cross-sectional study on functioning and HRQOL of children with a CHD. We recently reported on the HRQOL scores and the determinants of HRQOL of the children in a cohort study.[3] The subjects were 10- to 14-year-old children with a CHD treated at our hospital and their parents. We were particularly interested in the parent-child agreement in this age range because in the Netherlands there is a transition from primary school to secondary school in this age range and parental control decreases when children attend secondary school. Also, the average onset of puberty is at this age when children start sharing less information with their parents, which may also affect agreement. Children were excluded if they had a mental or developmental delay or insufficient knowledge of the Dutch language.

From this sample, we randomly selected 300 subjects using a computer-generated random sequence and we stopped the inclusion when we reached the amount of 120 participants (response rate of 40%). We found no differences between participants and nonparticipants regarding sex, diagnosis, and severity of the CHD.

The local ethics committee approved the study, and children above 12 years of age and all parents gave their informed consent to participate.

Participants

The questionnaires on HRQOL were completed by 115 of the children of whom 106 had a corresponding parent report. In this article, parents of adoptive children are also referred to as parents and therefore all parent-reports were filled out by 1 of the child's parents. Patient characteristics were administered and each child's medical diagnosis was registered according to the International Federation of Societies for Surgery of the Hand classification system.[8] We expressed severity of the CHD by means of bilateral involvement, number of affected digits per hand, and comorbidity. Comorbidity was defined as the presence of any comorbidity not related to the hand problem, or the presence of syndromal differences related to the hand problem but in different body parts (eg, esophageal atresia, cardiac problems).

HRQOL Measure

HRQOL was assessed by means of a generic questionnaire, Pediatric Quality of Life Inventory (PedsQL) 4.0 generic core scales, in Dutch, which has been proven to be reliable and valid.[9] The PedsQL consists of 23 items and 4 generic core scales: physical health (8), emotional functioning (5), social functioning (5), and school functioning (5). The psychosocial health score is calculated from emotional, social, and school functioning scores, and the total score is an average of the scores on all 4 generic core scales. A 5-point Likert scale is used to answer the questions (0=never a problem, 1=almost never a problem, 2=sometimes a problem, 3=often a problem, and 4=almost always a problem).[9] Each answer is reversed and rescaled on a 0 to 100 scale (0=100, 1=75, 2=50, 3=25, and 4=0), so higher scores indicate better HRQOL. Parent-reports and child self-reports are of parallel content.

We used 2 different age versions of the PedsQL: for ages 10 to 12 years, parents and their children filled out parent-report and child self-report for ages 8 to 12 years. For age 13 to 14 years, children and their parents filled out reports for ages 13 to 18 years.

Determinant Measurement: Covariates

Subject characteristics that were determined as possible covariates for child-parent agreement were the child's sex, age, and ethnicity. Three ethnic groups were made based on the country of birth of the children and their parents: Dutch, foreign, and adoptive. Other variables that were taken into account as possible covariates of the child-parent agreement were unilateral or bilateral involvement, number of affected digits per hand, and comorbidity (yes/no).

Statistical Analysis

Levels of total HRQOL and subdomains (physical health, emotional functioning, social functioning, school functioning, and psychosocial functioning) were calculated for children and their parents. In order to compare differences between child self-report and parent-report, mean scores and SDs were summarized separately.

To assess whether children and their parents agreed on the level of HRQOL, we examined the relation between scores of parents and children on different levels. To determine whether the children's and parent's responses differed significantly when comparing the means of the groups, we performed a 2-tailed, paired-samples t test at a criterion level of $P<.05$.

Agreement was also determined using intraclass correlation coefficients (ICCs). The ICC was estimated by a 2-way random effects model as a ratio of between-child/parent to total variance, where total variance includes variation between child/parent and within child/parent.[10-12]

We computed the means of the absolute within child/parent differences, the means of the differences (mean bias), and the SD of differences to assess the magnitude and range of individual differences between children's and parents' responses.[13] In this calculation, a mean bias smaller than zero indicates that parents score higher than their children and a mean bias greater than zero indicates that children score higher than their parents.[13] Consecutively, the graphical method of Bland and Altman[13] plots was used to illustrate the differences in responses pairwise. Limits of agreement were calculated as mean bias $\pm$ 1.96 times the SD of the difference. Mean bias and the limits of agreement levels were drawn as horizontal lines in the scatter plot. The Bland and Altman plot demonstrates not only the overall degree of agreement, but also whether the agreement is related to the underlying value of the item. For instance, parents of children with a low quality of life score may agree more closely with their children than parents of children with higher scores.

Linear regression models were used to assess the relationship of each independent variable of interest with agreement on both the PedsQL total score and subdomain scores. Statistical significance was set at $\alpha=.05$. Linear regression model assumptions were examined and satisfied. All data were analyzed using SPSS for Windows version 17.0.[a]

RESULTS

Demographics

The demographic characteristics of the 106 children participating in the study are presented in table 1. The 5 most common diagnoses were: radial polydactyly (16%), symbrachydactyly (9%), aplasia (8%), and hypoplasia, including longitudinal radial deficiency (8%) and syndactyly (5%). The syndromes that were found 3 times or more were VATER/VACTERL association (vertebral defects, anal defects, cardiac defects, esophageal defects, renal defects, limb defects), EEC syndrome (ectrodactyly-ectodermal dysplasia-clefting syndrome), Poland syndrome, and constriction ring syndrome.