Post-operative quality of life in children with severe Perthes disease: differences to matched controls and correlation with clinical function

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Abstract

The diagnosis of Legg-Calvé-Perthes disease (LCPD) has a considerable influence on the daily life of the patients with restrictions in their leisure time activities. This might influence their mood. Until now this aspect of the disease has been neglected. Therefore the objective of the study was to evaluate the health related quality of life (HRQoL) of children with severe LCPD who had an extensive surgery with pelvic/femoral osteotomy. The KIDSCREEN-10 and the modified Modified Harris Hip Score (mHHS) -questionnaire were administered to 17 children (16 boys and 1 girl) aged 5 to 11 years at the time of surgery. Analyses of mHHS were made preoperatively and at the time of the follow-up examination at least 2 years postoperatively. KIDSCREEN-analyses were made postoperatively. The follow-up results were compared to an age-matched normal control group. Correlations were computed between KIDSCREEN-10 and mHHS pre- and post-operatively. The postoperative calculated KIDSCREEN-10-T-value [70.2 (SD 12.7)] was higher than the mean T-value of the control-group [56.6 (SD 10.4)]. The mHHS improved from 54.4 (SD 19.9) to a score of 99.5 (SD 1.5) postoperatively. A strong correlation was found between the preoperative mHHS and the postoperative KIDSCREEN-10-T-value (Spearman's-rho 0.67, P=0.003). After containment improving surgery and a mean follow-up period of 4.2 years the HRQoL-status is even better compared with a healthy age-matched control group. As well an excellent clinical function could be achieved.

Introduction

Legg-Calvé-Perthes disease (LCPD) is a complex pediatric hip disorder which usually affects children in primary school age. The course of the disease may last for several years. In the early stage children often complain about discomfort or pain especially after physical activities.[1,2]

After diagnosis behavioral recommendations for daily life include reduction of physical activities, especially sport disciplines which produce a huge peak impact load for the joint.[2] Most of the children receive physiotherapy over several months for improving joint mobility; some get an orthosis prescribed either to keep containment or to reduce loading of the joint although a positive effect has not been proven.[3,4] In case of loss of containment surgical procedures like femoral or/and pelvic osteotomies are performed.[5] All these factors restrict the children in their daily life, in their leisure time activities and in the interaction with their peer group. These restrictions may influence their mental and social development. Until now this aspect of the disease has not been sufficiently considered in research. Therefore the objective of this study was to evaluate the *health related quality of life* (HRQoL) of children with LCPD. Due to the fact that the individual course of the disease and the treatment concepts differ considerably, we chose a group of patients who were severely affected and had an extensive surgery with pelvic (Salter or Triple) and femoral osteotomy.

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Key words: Legg-Calvé-Perthes disease, osteotomy, KIDSCREEN, quality of life, modified Harris Hip Score.

Acknowledgments: the authors would thank all children and parents that participated at the study

Contributions: NKP, first author; NKP, BW, CZ, clinical consultation; DR, statistical evaluation; all authors, study design.

Conflict of interests: the authors declare no potential conflict of interests.

Received for publication: 21 August 2014.
Accepted for publication: 7 September 2014.

Orthopedic Reviews 2014; 6:5567
doi:10.4081/or.2014.5567

Materials and Methods

Legg-Calvé-Perthes disease patients and control subjects

In the context of a LCPD-outcome study 17 children were recruited for the HRQoL-aspect of this study. Inclusion criteria were unilateral hip involvement, a severe course of the disease with Caterall type 3 or 4, Herring B or C and a loss of containment demanding a pelvic and femoral osteotomy (Figure 1).

The minimum follow-up time was 2 years postoperatively. Exclusion criterion was an additional disease that might affect HRQoL over a certain period of time. All children had to attend a physiotherapy program over at least 1 year postoperatively. In addition they were ordered to reduce their sports activities to avoid impact load and were encouraged to swim and cycle after consolidation of the osteotomies.

In the period from 2002 to 2008, 43 patients were treated by a pelvic and femoral osteotomy for severe LCPD by two surgeons, 24 fulfilled the inclusion criteria; 17 patients (16 boys, 1 girl) could be recruited - corresponding follow-up-rate 70.8%. The mean age at the time of surgery was 8.0 years (SD 1.7, range 5.1-11.3 ys.) and the mean follow-up time was 4.2 years (SD 1.8, range 2.0-7.5 ys.). At the time of follow-up the mean age was 12.1 years (SD 2.1, range 8.3-15.3 ys.).

For the comparison of the follow-up results 63 healthy children (24 girls, 39 boys) from the community with a mean age of 12.1 years (SD 2.0, range 8.0-15.8 ys.) were recruited as an age-matched normal control group to define a normal HRQoL-score. The controls were chosen randomly from a heterogeneous pool with respect to socioeconomic status; this status was not controlled.

The study protocol was approved by the local ethics committee and all children and parents gave their written informed consent.

KIDSCREEN-10 score

For evaluating the HRQoL the *KIDSCREEN-10 score* was chosen as an appropriate measuring instrument. It consists of 10 items and each question has to be answered on a 5-point response scale. These are the questions of KIDSCREEN-10 questionnaire:[6] i) Have you felt fit and well? ii) Have you felt full of energy? iii) Have you felt sad? iv) Have you felt lonely? v) Have you had enough time for yourself? vi) Have you been able to do the things that you want to do in your free time? vii) Have your parent(s) treated you fairly? viii) Have you had fun with your friends? ix) Have you got on well at school? x) 10. Have you been able to pay

attention?

KIDSCREEN-10-Data were collected postoperative at the time of the follow-up examination at least 2 years postoperatively. The scoring of the KIDSCREEN questionnaires was carried out based on the guidelines of the European KIDSCREEN Group. The validity and reliability of KIDSCREEN have been proven in the European population. The answers were coded in numbers and summed for each child. Rasch person parameters were assigned to each sum score and were transformed into T-values as documented in the scoring model of the KIDSCREEN handbook.[6,7] A low score implicates a poor, a high score a better HRQoL.[8]

Modified Harris Hip score

To correlate the HRQoL with the clinical situation the *modified Harris Hip Score* (mHHS) was analyzed pre- and post-operatively. The mHHS includes an assessment based on *pain* (maximum 44 points) and *function* (maximum 47 points). The elements of *deformity* (maximum 4 points) and *range of motion* (maximum 5 points) from the original Harris Hip Score are excluded.[9] The total sum score was multiplied for every child with 1.1, so that a total possible score of 100 points could be reached; a score of 90 to 100 pts. means an excellent, 80 to 89 pts. a good, 70 to 79 pts. a fair and less than 70 pts. a poor result.[10,11]

Statistical analysis

Descriptive statistics were computed for all variables of the KIDSCREEN-10 survey postoperatively and for the mHHS at two points of time: t_0 preoperatively and t_1 postoperatively. They were all reported as means and standard deviations (SD). Rasch scores and T-values were calculated according to the KIDSCREEN-10 manual.[6] The independent samples t-test was used for the comparison of normal control group KIDSCREEN-T-value with the postoperative T-value of patient collective, as well as differences in individual KIDSCREEN-items between control group and patients group.

Correlations were computed between the HRQoL (KIDSCREEN-10) and the clinical result (mHHS). They were based on the Spearman`s rho for the T-values and Kendall-tau-b correlation coefficient for individual KIDSCREEN-10 items as non-parametric measures of statistical dependence. A $P<0.05$ was considered statistically significant. The Bonferroni-Holm-procedure was applied to the comparisons between patients and the control group, to keep the number of false-positive results low. Power-sample-size computation yielded, that a small effect of size 0.31 (Cohen's d) can be detected in the comparisons between controls and patients, for the given alpha-level and desired statistical power of 0.8. All statistical analyses were conducted using SPSS Version 21.0.

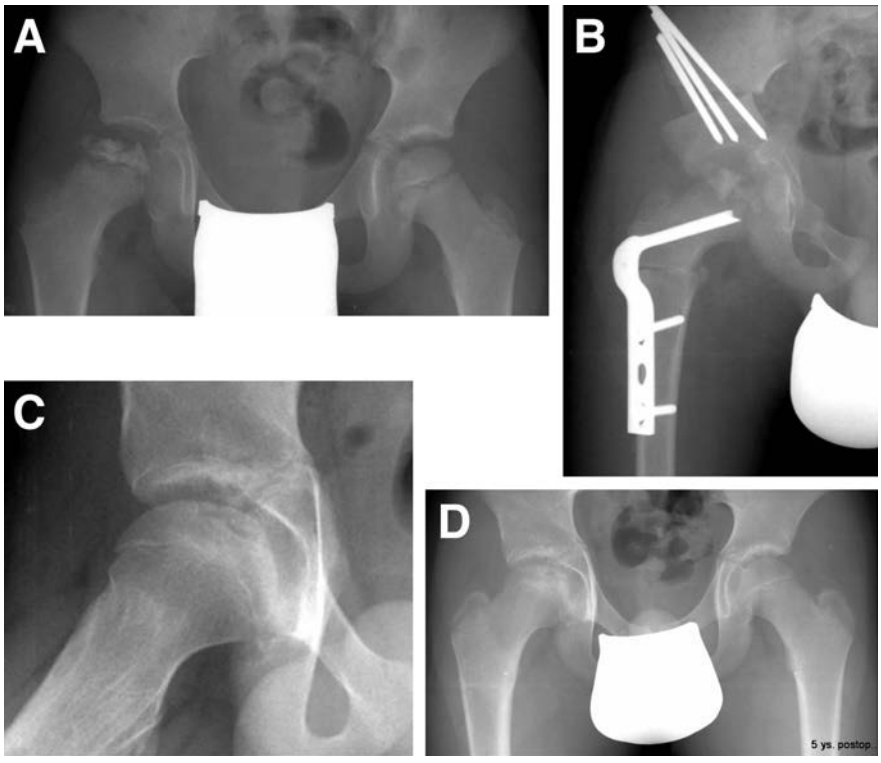

Figure 1. A 7-year-old boy with severe Legg-Calvé-Perthes disease and loss of containment (A); (B) X-ray after containment improving surgery with triple and varus/rotation femoral osteotomy; (C,D) 5 ys. postoperatively.

Results

Complete mHHS and KIDSCREEN-10 results were available for all 17 patients. The postoperative mean KIDSCREEN-10-score-T-value was calculated at 70.2 (SD 12.7, range 53.1-83.8).

The age-matched control group achieved a mean KIDSCREEN-10-score-T-value of 56.6 (SD 10.4, range 38.3-83.8); the comparison with the postoperative T-value of children with LCPD (Figure 2A) revealed a significant difference indicating a higher HRQoL-Level in LCPD-patients after surgery ($P<0.001$).

Detailed analysis revealed that LCPD-patients reported that they felt more comfortable ($P<0.001$) and powerful ($P<0.001$) in comparison to the healthy control group (Figure 3). Moreover, the psychological well-being was significantly higher: patients stated to feel less sad ($P=0.002$) (Figure 3). The *time for themselves* and the organization of their leisure time activities was also significantly higher ($P<0.001$) (Figure 3). Furthermore the LCPD-patients stated to concentrate better at school than the normal control group ($P=0.003$) (Figure 3).

The mean mHHS increased from 54.4 (SD 19.9) at t_0 to a score of 99.5 (SD 1.5) at t_1 (Figure 2B). While preoperatively 11 (65%) children demonstrated a poor mHHS of less than 70 pts. and the remaining 6 a fair score (mHHS: 70 to 80), after a mean follow-up of 4.2 ys. postoperatively all patients had excellent results with a mHHS of more than 90 pts. This was statistically highly significant ($P<0.001$).

The analysis of the correlations between mHHS and KIDSCREEN-10 resulted in a strong correlation between the preoperative (t_0) mHHS and the postoperative (t_1) KIDSCREEN-10-T-value (Spearman's-rho 0.67, $P=0.003$). In addition we found significant correlations between the mHHS at t_0 and the subscales of KIDSCREEN-10 at t_1: they were statistically significant for the KIDSCREEN-10 subscale *feeling fit and well* at t_1 (Kendall-tau-b 0.46, $P=0.032$) as well as for the subscale *being able to pay attention* at t_1 (Kendall-tau-b 0.47, $P=0.027$). No correlation was found between postoperative mHHS and postoperative KIDSCREEN-10-index-score.

Discussion and Conclusions

Up to now the HRQoL of children with LCPD has not been sufficiently subject to research projects although this diagnosis, the consecu-

tive behavioral restrictions and the treatment modalities implicate a severe impact on their daily life. Therefore the aim of the present study was to evaluate the HRQoL after extensive surgical containment treatment. As measuring-instrument the KIDSCREEN-10-questionnaire was chosen, a validated and established test.[7,8,12] The results were correlated with the functional status measured by the mHHS.

As a result the study showed that on average 4.2 years after complex surgery an excellent clinical result could be achieved. The evaluation of the HRQoL also resulted in very positive findings: the LCPD patients achieved a significantly higher KIDSCREEN index score compared to the age-matched healthy control group and as well as in comparison to a historical healthy German control group aged 8-18 ys. [52.1 (SD 9.5)].[6]

These positive findings demonstrate that after LCPD – even with a severe course – HRQoL is not impaired – it is quite the contrary: HRQoL is significantly higher. A possible explanation might be that children with a disease like LCPD which required extensive therapy and limited their daily life during a long period appreciate more their regained health and daily activity options and therefore better assess their HRQoL in the KIDSCREEN questionnaire.

In further studies it would be interesting to calculate as well the preoperative HRQoL for the comparison with the postoperative result. In this study it was not possible to acquire preoperative KIDSCREEN-Data, because KIDSCREEN-questionnaires exist only since 2006.[6]

The correlation analysis pointed out that children who had a better preoperative mHHS and therefore a better clinical function showed a higher HRQoL after surgery. This is also underlined by the correlation results of preoperative mHHS with the subscales of postoperative KIDSCREEN-10: Children with better preoperative clinical function felt postoperative more comfortable and fit, as well as paying more attention at school. Surprisingly no correlation could be found between postoperative mHHS and HRQoL. This might be explained by the small study population and the excellent results: there is not a single patient with a poor or fair clinical outcome and the T-scores are on average significantly better than these of a healthy control group. Of course further studies with a bigger study population are necessary to confirm these findings.

The present study is to our knowledge the first attempt to determine HRQoL of children with severe LCPD. Up to now there has been only two studies looking for behavioural sequelae of LCPD: Price et. al evaluated patients who were treated either by an orthosis or by surgery, by the Louisville Behavioral Checklist; as a result the bracing group was more likely to demonstrate deficits in social, academic and sexual behavior as compared with patients in the surgery group.[13] The opposite was found by Khurana *et al.* who concluded that LCPD treated non-operatively using abduction cast and brace did not affect the emotional and mental well-being of patients in the longterm.[14]

Therefore we feel that the evaluation of the HRQoL-aspect is worthwhile because parents are deeply concerned if they get the diagnosis of LCPD for their child, hear the recommendations for the daily life (restrictions for sport activities, intensive physiotherapy, in some cases hip unloading by using crutches or a wheelchair etc.) and finally get in severe cases the recommendation for a surgical procedure.

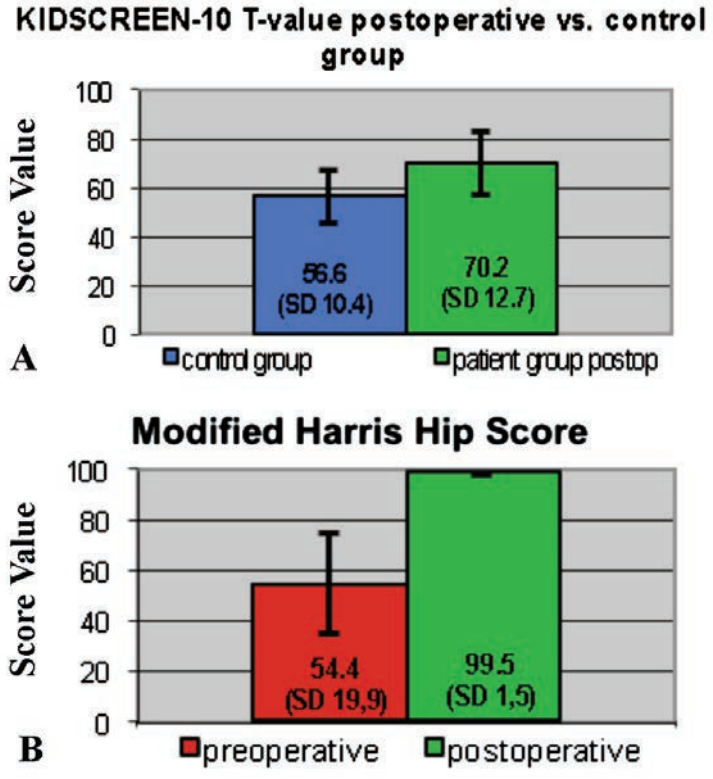

Figure 2. A) Comparison of mean and standard deviation of postoperative KIDSCREEN-10 score *vs.* conrol group. B) Comparison of mean and standard deviation mHHS pre- and postoperatively.

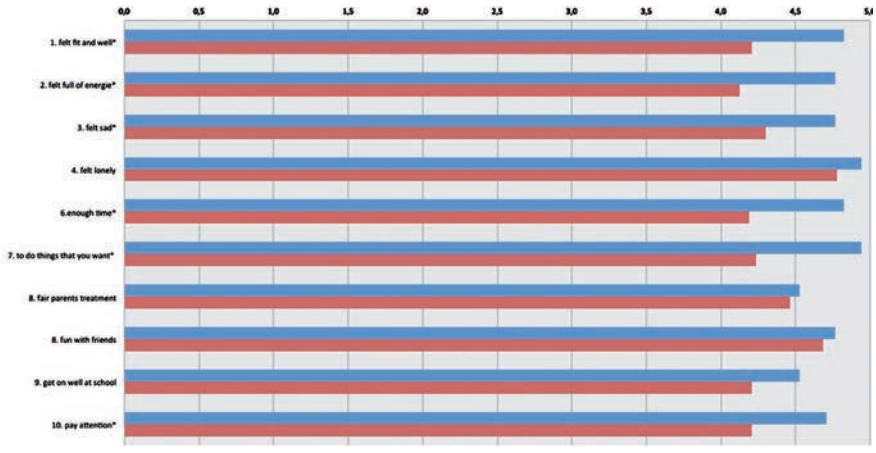

Figure 3. Results of the KIDSCREEN 10-questionnaire: patient group postoperative (blue) *vs.* normal group (red). Asterisks indicate significant values.

But children are usually at an age, in which motor activities with their peers play an important role in their daily life. Therefore parents understandably also worry about the psychosocial development of their child.

In conclusion this study is the first to demonstrate that patients with severe LCPD do not have any impaired HRQoL after a follow-up period of at least 2 ys. after containment improving surgery; the quality of life status is even better compared with a healthy age-matched control group. Therefore, worried parents can be calmed, their children will return to a normal attitude towards life.

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