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Quality of Life in Pediatric Patients with Avascular Necrosis of the Femoral Head

Luanne Lísle dos Santos Silva[1(A,B,D,E,F)], Marilda Castelar[1(A,D,E,F)], Marcos Almeida Matos[1(A,C,D,E,F)]

Bahiana School of Medicine and Public Healt, Salvador, Brazil

SUMMARY

Background. The aim of the present study was to evaluate health-related quality of life in pediatric patients with avascular necrosis of the femoral head.

Material and methods. A comparative study was conducted involving 32 children and adolescents in "ONFH Group" (subjects with a diagnosis of ONFH) and 44 in "Comparison Group" (asymptomatic children), both aged between 8 and 18. Clinical and sociodemographic data were obtained along with PedsQL 4.0 questionnaires to evaluate the quality of life in the age-ranges studied.

Results. The two groups were similar as regards sociodemographic characteristics. Comparison of the quality of life total scores demonstrated significant differences between the ONFH group and the comparison group (58.86±16.54 versus 82.08±9.51), and this also occurred in all the domains analyzed. The predictors that showed correlation with the quality of life were: presence of necrosis, age, time of diagnosis, radiographic classification, Charnley score, educational level, previous surgeries, and previous infections.

Conclusion. Our data confirm the hypothesis that pediatric patients with ONFH demonstrate a lower quality of life than healthy children in all the domains evaluated, especially with regard to the physical aspect.

Key-words: osteonecrosis; quality of life; hip

BACKGROUND

Osteonecrosis of the femoral head (ONFH) is a progressive disorder that mainly affects young persons. It is characterized by diminished vascular supply to the femur leading to collapse of the subchondral bone and secondary degenerative hip arthropathy in up to 80% of cases [1,2,3]. Pain and functional limitations resulting from ONFH are often mentioned in the literature, but they are always described in adults or young adults [1,2,3]. However, because of chronic suffering, ONFH has all the characteristics that could lead to it having strong impact on the health and quality of life of pediatric patients.

Evaluating the quality of patients' health based almost exclusively on the clinical severity of the disease has certainly created distortions between the medical approach and expectation of individuals, especially in chronic diseases such as ONFH [4]. Health-related quality of life (HRQoL) is a concept that may provide the subject's own view of his/her well being and not the view of a health professional about the disease [4,5]. Evaluation of health-related quality of life (HRQoL) has, for this reason, become increasingly important in clinical practice as the main way of approximating medical practices and patients' expectations.

In spite of the importance of HRQoL in medical evaluation, few studies have focused on ONFH, and rarer still are those that were dedicated to diseases in the pediatric age-range [6,7,8,9]. For this reason, the aim of the present study was to evaluate health-related quality of life in pediatric patients with avascular necrosis of the femoral head.

MATERIAL AND METHODS

A comparative study was conducted at the outpatient Pediatric Orthopedic Clinic of the Hospital. The affected group was composed of patients with a previous radiographic diagnosis of ONFH (ONFH group), while the non-affected group (comparison group) were asymptomatic individuals from a public school of the same city. The research was conducted in the period from September 2012 to October 2014, after the study had been approved by the Ethics Committee of the Institution and the parents or guardians of the individuals had signed the Informed Consent Term.

For the ONFH group, children with osteonecrosis of the femoral head according to Bucholz-Ogden radiographic criteria [6] aged between 8 and 18 years were included and subjects who did not have adequate cognition and those with neurological sequelae, and other diseases of relevance for the musculoskeletal system were excluded. For the comparison group, healthy children between the ages of 8 and 18 whose socio-demographic characteristics were similar to those of the subjects from the ONFH group were selected; all the individuals underwent clinical orthopedic evaluations to guarantee the absence of signs and symptoms of hip dysfunction; for ethical reasons, no radiographic evaluations were made.

Based on Hailer et al. 2014, the estimated sample size was 34 subjects for each group, calculated to detect a difference of 10%, considering the prevalence of changes in the quality of life of 20%, alpha error of 5%, with a test power of 90% and estimated loss of 10%. There were a total of 32 children in the ONFH group and 44 in the comparison group.

After selection and inclusion of all the children participating in the study, clinical and sociodemographic data were obtained, such as age, gender, weight, height, race, origin, educational level (in years of schooling), family income, etc. In addition, the following questionnaires were applied: PedsQL 4.0 to evaluate the quality of life in the age-ranges studied [5,10], and the hip score modified by Charnley, to evaluate hip dysfunction [11]. The Charnley score was used for quantitative and qualitative evaluation of the state of the hip articulation. This score evaluates the arch of movement, level of pain and walking, totaling 18 points in a clinically normal hip, and with lower scores indicating dysfunction of the hip. Although the Charnley score was designed for adults, it has also been efficiently tested in the pediatric population [3,12].

The PedsQL 4.0 was used after obtaining authorization from the *Research Trust* in the Brazilian Portuguese version specifically developed for the child population, with defined and validated intervals [5, 10]. The PedsQL 4.0 patient's form can be filled out by research subjects themselves, but as an option, filling out and data collection was performed in the form of an interview, due to the low educational level expected for the two groups [5,10]. The responses were reclassified into numerical values and each domain score was an arithmetic mean of the question scores. In this scoring system, higher scores indicate better quality of life [5,10].

For statistical analysis, descriptive presentation of the continuous data in the form of means and standard deviations was used, while the discrete variables were presented as frequencies. For comparison of the variables between the groups, the Chi-square test was used for the discrete variables, and the t-test for continuous variables. When factors associated with the quality of life were identified in the univariate analysis, multivariate analysis was performed in search

of confounding factors. All analyses assumed a p value of less than 0.05 as the significance threshold.

RESULTS

The final sample was composed of 32 children and adolescents in the ONFH group and 44 in the comparison group. The statistical power of the sample was 98%, taking into consideration an alpha error of 0.05 and difference of 10 points to be detected in quality of life. The mean age was 11.58 ± 3.20 years, with 48 (63.2%) subjects being in the age-range of children and 28 (36.8%) in the age-range of adolescents. The sociodemographic and clinical characteristics of the two groups are shown in Tab. 1. Comparison of the quality of life scores (PedsQL.4.0) demonstrated differences in all the domains analyzed (Tab. 2).

The predictors that correlated with the quality of life were: ONFH, age, time of diagnosis, radiographic classification, Charnley score, educational level, previous surgeries, and previous infections. After the multivariate analysis, the only factor that remained

Tab. 1. Sociodemographic and clinical characteristics of both groups

Characteristics	ONFH Group % or mean (± SD)	Comparison Group % or mean (± SD)	p
Age*	12.81 (±3.62)	10.68 (±2.54)	0.006
Age group**			0.074
8-12	16 (50%)	32 (72.3%)	
13-18	16 (50%)	12 (27.2%)	
Weight	43.91 (±13.51)	40.14 (±13.72)	0.238
Height	1.51 (±0.16)	1.47 (±0.15)	0.229
BMI	19.04 (±4.51)	18.14 (±3.35)	0.325
Gender (male%)	19 (59.37%)	28 (63.63%)	0.706
Race			0.712
White (%)	2 (6.25%)	4 (9.09%)	
Mulatto (%)	17 (53.12%)	26 (59.09%)	
Black (%)	13 (40.62%)	14 (31.82%)	
Scholarship (years)	5.85 (±2.51)	4.26 (±1.89)	0.08
Characteristics	ONFH Group n, % or mean (± SD)	Comparison Group n, % or mean (± SD)	p
Time of diagnosis (months)	12.41 (±23.83)	-	-
Trauma history	11 (34.37%)	2 (4.54%)	0.001
Infection in the last year (n)	12	1	0.001
Previous surgery	23	8	0.001
Number of surgeries (n)	1.19 (±1.33)	0.21 (±0.46)	0.001
Hospitalization	0.56 (±0.98)	0.05 (±0.21)	0.006
Chronic use of medication	13	1	0.001
Risk factors		-	-
Trauma	1		
Sickle cell disease	9		
Septic arthritis	5		
Corticosteroid therapy	1		
Hip dysplasia	3		
Epiphysiolysis	4		
Fracture	1		
Perthes' disease	7		
Necrosis classification		-	-
I (%)	9 (28.12%)		
II (%)	7 (21.87)		
III (%)	5 (15.62%)		
IV (%)	11 (34.37%)		

SD – standard deviation; *Continuous variable; **categorical variable

Tab. 2. Comparison of quality of life of both groups according to PedsQL domains

Variable	Group ONFH mean (± sd)	Group Comparison mean (± sd)	P
PedsQL total	58.86 (±16.54)	82.08 (±9.51)	0.001
Physical Functioning	53.91 (±24.44)	88.77 (±7.27)	0.001
Emotional Funcioning	60.78 (±21.59)	6.,48 (±14.89)	0.204
Social Functioning	70.31 (±19.71)	90.68 (±12.56)	0.001
School Functioning	59.06 (±23.81)	78.75 (±16.04)	0.001

as an independent predictor of the quality of life was the Charnley score, accounting for 70.18% of the score (p=0.001). When the Charnley score was removed from the model, the presence of necrosis became the only significant predictor, accounting for 72.2% of the score (p = 0.007). Following removal of the presence of necrosis, the radiographic classification began to account for 50.9% of the score (p = 0.001).

DISCUSSION

The quality of life of children with avascular necrosis of the femoral head measured by PedsQL.4.0 was significantly lower (58.7 points) when compared with the quality of life of the control children (82.1 points). This loss was demonstrated in all the domains evaluated by the PedsQL 4.0. The greatest harm to HRQoL occurred in the domain of physical capacity (53.9 versus 88.8) and the lowest, in the emotional domain (60.7 versus 66.5).

In the univariate analysis, the presence of ONFH, age, time of diagnosis, radiographic classification, Charnley score, educational level, previous surgeries, and previous infections were the predictive factors associated with a decrease in the quality of life. The multivariate analysis, however, demonstrated that ONFH was the only factor independently associated with loss of the quality of life of the patients in all the models tested.

Pediatric patients with chronic diseases normally have a consistently lower quality of life than that of the healthy child population. ONFH is a chronic condition that severely affects hip function, leading to physical limitation, pain and prolonged treatment [12]. These findings are consistent with those studies that have shown pain to be directly associated with a reduction in quality of life of the pediatric population in general and also with the fact that the physical capacity domain was the one most affected in our patients [12,13,14].

Dale et al.[4] evaluated the quality of life in 127 children with sickle cell anemia and found an overall PedsQL 4.0 score of 68.6 points. The etiology of ONFH is also associated with chronic diseases, such as sickle cell disease, septic arthritis, developmental dysplasia of the hip, and epiphysiolysis, which have been shown to be capable of producing loss of quality of life [2,3,6,12,13]. In these diseases, children undergo multiple clinical and surgical treatments, the results of which may be accentuated loss of function, dissatisfaction and physical, financial and emotional exhaustion [2,3,6,12,13].

In the current study, the severity of ONFH was maximum in 34.45% of the cases (Bucholz-Ogden Grade IV) and the participants presented important limitations of hip function associated with pain, as demonstrated by a Charnley score of 15.8 points in the ONFH group. Twelve patients had had infection in the past year and 23 had undergone surgical procedures. These data emphasize the human suffering of the study population accounting for the remarkable loss of quality of life. Our patients were shown to have a lower quality of life than children with sickle cell disease, and this fact emphasizes the potentially devastating role that osteonecrosis could have when it appears during the course of a chronic disease, especially as it affects the physical capacity of individuals.

Our findings also point towards the confirmation that ONFH alone is capable of producing loss of quality of life, even in cases regarded as milder. The participants who had ONFH and associated sickle cell disease were those who had the worst HRQoL scores; however, significantly lower scores were also found in patients with Perthes' disease, which is traditionally considered "silent" or asymptomatic after its acute stage. The multivariate analysis also demonstrated that irrespective of other associated factors (including the etiology), ONFH was an independent predictor for loss of HRQoL.

As far as we know, there is no similar study of ONFH in pediatric patients that could serve as comparison. Hailer et al.[9] 2014, conducted a study with patients who had Perthes' disease. In spite of this study having been conducted in adults, the authors confirmed a lower quality of life in patients with Perthes' disease in comparison with the general population, especially in the domains of mobility, usual activities, and pain.

Our study was subject to some limitations. The study evaluated patients with ONFH suffering from multiple etiologies and should be regarded as a pilot report for the design of larger studies that would focus on specific disorders such as Perthes' or sickle cells disease. Despite the limitations, the etiology did not independently affect quality of life and the results of the current study help to understand the important impact that ONFH has on the quality of life of pediatric patients.

Our study represents an original contribution to ONFH in children, especially from the point of view of medical evaluation based on the perception of the subject. Our data allow us to confirm the hypothesis that pediatric patients with ONFH present a lower quality of life than healthy children in all the domains evaluated, especially with regard to the physical aspect. Secondarily, predictive factors for loss of quality of life in these patients were also identified, namely, hip function, time of diagnosis, radiographic severity of the lesion, educational level, and previous surgeries and infections.

CONCLUSION

Our data confirm the hypothesis that pediatric patients with ONFH demonstrate a lower quality of life than healthy children in all the domains evaluated, especially with regard to the physical aspect.

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Adres do korespondencji / Address for correspondence
Prof. Marcos Almeida Matos
Rua da Ilha, 378, Itapuã, Salvador-Bahia, Brazil, 41620-620
Telefax (55) 71-3358-8886, e-mail: marcos.almeida@hotmail.com

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