HEALTH-RELATED QUALITY OF LIFE IN HEARING-IMPAIRED CHILDREN AND ADOLESCENTS IN MONTENEGRO

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ABSTRACT

The aim was to assess the health-related quality of life (HRQoL) of Montenegrin children with hearing aids (HA) or cochlear implants (CI) at school age and to analyze parental views of their children HRQoL. The total number of children and parents who participated was as follows: 25 children with HA and 38 parents, 19 children with CI and 26 parents, and 30 children with normal hearing and 31 parents. Children and parents were surveyed with the KINDL-R instrument. The children with HA reported significantly worse HRQoL than the NH group in overall HRQoL score, in everyday functioning, family, and friends subscales. The difference between the children with CI and NH was nearly statistically significant (p=0.099). Parents estimated that children with NH had significantly better HRQoL in the total mean and physical well-being, emotional well-being, self-esteem and friends subscales than children with HA. We found statistically significant positive correlations between parents' and children's responses on the family and everyday functioning subscales for the HA and CI group. The children with HA assessed HRQoL similarly to children with CI and significantly lower than NH children. Children with CI achieved similar HRQoL to NH peers.

Key words: Cochlear implant; Hearing aid; Hearing impairment; KINDL-R; Quality of life

INTRODUCTION

Sensorineural hearing impairment (HI) is expected in the pediatric population, with a rate of 1-6 per 1000 children [1] and affects 34 million children worldwide [2]. Hearing loss often causes difficulties not only for the children but also for the family. HI children are at high risk for deficits in language expression, poorer interpersonal communication, reduced social interactions, poorer academic performance, psychosocial well-being, and long-term quality of life (QoL). Health-related quality of life (HRQoL) is a subjective, multidimensional measure defined as an individual's perception of the impact of illness and treatment on QoL [3,4]. At a minimum, it includes physical, mental, and social functioning. Individual HRQoL can only be assessed through subjective measures by interviewing the patient himself or, if the patient cannot provide a coherent answer, by interviewing a relative, friend, or close observer. Several generic and disease-specific QoL instruments have been used to evaluate pediatric hearing disorders. The generic instruments allow comparisons with healthy populations but may lack specificity and sensitivity to assess specific conditions or hearing interventions. The KINDL-R Questionnaire for measuring HRQoL in children and adolescents is a reliable, international, validated, and established generic pediatric health-related HRQoL questionnaire [5]. Early auditory rehabilitation through hearing aids (HA) and cochlear implants (CI) is critical for children with varying hearing loss and en-

ables integration into the hearing world.

Several studies have shown that pediatric CI users have a lower QoL than their healthy counterparts [6,7].

In contrast, other studies report significant improvement in HRQoL in children with CI [8-10] and similar quality of life to healthy children [11,12]. Children and adolescents who use HA perform worse than their normal-hearing peers in social acceptance, peer relationships, self-esteem, emotional adjustment, and communication [13,14]. The impact of unilateral and bilateral sensorineural hearing loss on children's QoL remains unclear.

In Montenegro, cochlear implantation was started in 2008. In the 15 years it has been used in Montenegro, 52 CIs have been performed in children. There are 26 patients with CI aged from 7 to 17 years. There are no published studies on HRQoL of children and adolescents with HA or CI in Montenegro in the available literature. Studying the HRQoL of children with HA or CI is essential to help policymakers, physicians, parents, etc., make decisions about funding, access to treatment, and selection of appropriate hearing interventions.

The present study aimed to assess the HRQoL of Montenegrin children and adolescents with HA or CI at school age and to compare their results with those of their normal-hearing peers.

The second aim was to analyze parental views of their children with HA and CI and to investigate the agreement between parents' self-assessment and children's self-assessment of HRQoL.

MATERIALS AND METHODS

A case-control study assessed the HRQoL of children and adolescents using HA or CI.

Ethics

The study was approved by the Ethics Committee of the Clinical Center of Montenegro (Approval number: o8/183) and conducted following the Declaration of Helsinki. Written permission was obtained from the hospital management of the departments in question. Permission to use the KINDL-R questionnaires was obtained from their respective author (Ulrike Ravens-Sieberer) (ID number: 1686). Parents and children were informed about the purpose of the study. Each participant had to sign an informed consent and did so on behalf of their child.

Participants and data collection

All patients were diagnosed and treated at the Clinical Center of Montenegro in Podgorica or the General Hospital of Cetinje. The study involved HI children and adolescents divided into two groups: children using HA(s) and children using CI(s). All children had to be between 7 and 17 years old. Children with a HI had to have been wearing their hearing device (s) for at least six months. The only exclusion criterion was that the child was unable to complete the questionnaire due to a significant physical, cognitive, or developmental delay. Parents of children with HA or CI who participated in the study formed the parent group. Both parents potentially participated in this study.

The control group of NH children consisted of healthy children aged 7 to 17 years and their parents who volunteered to participate in the study. The inclusion criterion was normal hearing, as the child's parents reported. All children had normal, age-appropriate development and no cognitive problems. Children with chronic diseases and physical or mental disabilities were excluded from the control group.

Parents whose children met the inclusion criteria were contacted by telephone and mail contact using a clinical database of children or were approached during clinical consultations by the researchers to inform them about the purpose and procedures of the study. Parents who consented to participate were requested to complete the questionnaire independently. Data were collected anonymously and electronically over three months, from September 1, until December 1, 2022. The transformed data were entered into a database by a researcher, and then another researcher double-checked the data entry for all participants.

Thus, the total number of children and parents who participated in this study and completed the HRQoL questionnaire is as follows: 25 children with HA (s) and 38 parents, 19 children with CI(s) and 26 parents, and 30 NH children and 31 parents.

The questionnaire

The questionnaire consisted of two parts. The first part recorded sociodemographic and clinical characteristics of the child and family, such as age, sex, type of school attended, age of diagnosis of hearing loss, duration of hearing device experience, and laterality of hearing loss. In the second part, the influence of HI on the children's HRQoL was investigated.

Generic HRQoL measure

For the present study, an internationally established questionnaire, the KINDL-R, was used to assess generic HRQoL in children older than seven years and their parents [15]. The KINDL-R self-report questionnaire developed in Germany was translated into several languages. Due to the close similarity of the Serbian and Montenegrin languages, the adapted Serbian version of the KINDL-R was used: KidKINDL_children_7-13y_Serbian, KiddoKINDL_adolescents_14-17_Serbian, and Kid_KiddoKINDL_parents_7-17y_Serbian [16].

The Kid and Kiddo questionnaires for children and parents are similar and contain 24 questions on a five-point Likert scale (1 =«never», 2 =«rarely», 3 =«sometimes», 4 =«often», 5 =«all the time»). The

questionnaires are divided into six dimensions. including physical well-being, emotional well-being, self-esteem, family, friends, and daily functioning (school), and refer to how the child felt during the previous week. The six subscale scores are converted to a 100-point scale, where o is the minimum, and 100 is the maximum HRQoL score, and combined into a single score representing total HRQoL. It is based on SPSS.

Statistical analyses

Descriptive statistics for baseline demographic data included frequencies and percentages for categorical variables and mean, standard deviation (SD), and range for continuous variables. A two-sample t-test was used to analyze the continuous variables that showed normal distribution and compare mean values between multiple groups by variance analysis. Regarding categorical variables, comparisons between groups were made with Pearson's Chi-Square and Fisher's Exact Test. Spearman's correlation analyses were used to examine the relationship between continuous variables. The Kolmogorov- Smirnov test was used before statistical analysis to determine whether the data were normally distributed. The results were evaluated within a confidence interval of 95%, and statistical significance was set to p < 0.05. The

SPSS 26.0 for Windows (Chicago, IL, USA) was used for statistical analysis.

RESULTS

The HA group included 13 females (52%) and 12 males (48%) with a mean age of 12.4 years (SD 3.2 years, range 7.0-16.9 years). There were 7 (36.8%) girls and 12 (63.2%) boys among the children in the CI group. The mean age of the CI group was 11.5 years (SD 3.3 years, range 7.2-17.1 years). The group of NH children included 16 females (53.3%) and 14 males (46.7%) with a mean age of 12.6 years (SD 3.0 years, range 7.1-17.0 years). There was no statistically significant difference between the study (HA and CI) and the control (NH) groups in terms of age, gender, and number of siblings. The mean duration of HA use was 5.8 years (SD 3.0 years, range: 1.2-13.3 years), and the mean duration of CI use was 6.8 years (SD 2.7 years, range: 1.1-11.2 years). There were no statistically significant differences between the HA and CI groups for experience with their respective devices and type of school attended. Laterality of hearing device fitting was statistically significantly different between the HA and CI groups showing the HA group to be significantly bilaterally fitted than the CI group (p = 0.006). Participant demographic and audiological data are displayed in Table 1.

Table 1. Socio-de	mographic cha	aracteristics	s of the study J	oarticipant	S		
	Children	with HA	Children	with CI	NH ch	ildren	
Characteristics	Mean (SD)	Range	Mean (SD)	Range	Mean (SD)	Range	р
Age (years)	12.4 (3.2)	7.0- 16.9	11.5 (3.3)	7.2- 17.1	12.6 (3.0)	7.1- 17.0	0.417
Experience with device fitting (years)	5.8 (3.0)	1.2- 13.3	6.8 (2.7)	1.1- 11.2	/	/	0.267
	n	%	n	%	n	%	
Total number	25		19		30		
Gender							0.488a
Male	12	48.0	12	63.2	14	46.7	
Female	13	52.0	7	36.8	16	53.3	
Number of siblings							0.996
None	3	12.0	2	10.5	3	10.0	
10ľ 2	19	76.0	14	73.7	23	76.7	
3 and above	3	12.0	3	15.8	4	13.3	
Type of school							0.710a
Regular	19	76.0	16	84.2	30	100.0	

	Children	with HA	Children	with CI	NH chi	ldren	
Characteristics	Mean (SD)	Range	Mean (SD)	Range	Mean (SD)	Range	р
Hearing impaired school	6	24.0	3	15.8	/	/	
Laterality of hearing device fitting							0.00ба
Unilateral	13	52.0	18	94.7	/	/	
Bilateral	12	48.0	1	5.3	/	/	
Number of parents included in study	38		26		31		0.463
Mother	28	73.7	16	61.5	19	61.3	
Father	10	26.3	10	38.5	12	38.7	
Hearing loss in the family							0.139a
Yes	10	26.3	5	19.2	/	/	
No	28	73.7	19	73.1	/	/	
Unknown	0	0.0	2	7.7	/	/	

HA - Hearing aid, CI- Cochlear implant, NH- Normal hearing, SD- Standard deviation

a Compared HA and CI groups

HRQoL of children with HA and NH

The HRQoL total score on the KINDL questionnaire was 75.92 ± 6.55 for HA and 80.83 ± 7.33 for NH children (Table 2).

Table 2. Mean subsc	ales and total scores from the KINDL-R for children and parents (score/100)							
	Children	with HA	Children	n with CI	NH children			
Mean (SD)	Child	Parent(s)	Child	Parent(s)	Child	Parent(s)		
Physical well- being	83.25 (12.59)	79.44 (11.79)	85.85 (10.38)	81.73 (12.86)	84.17 (13.41)	89.72 (7.66)		
Emotional well- being	80.50 (12.93)	78.62 (12.47)	77.30 (12.88)	78.61 (14.16)	79.17 (13.37)	87.50 (7.74)		
Self-esteem	73.50 (9.42)	75.66 (9.72)	73.36 (9.04)	77.88 (11.07)	75.63 (12.32)	81.85 (8.12)		
Family	77.25 (13.24)	84.21 (9.49)	75.66 (12.99)	85.82 (12.56)	85.21 (9.78)	85.89 (7.73)		
Friends	72.75 (10.96)	81.25 (8.96)	81.25 (9.08)	82.93 (11.67)	86.25 (8.75)	86.69 (8.19)		
Every day (School) Functioning	68.25 (10.66)	76.48 (11.20)	70.07 (10.74)	78.36 (13.94)	74.59 (11.83)	81.45 (10.14)		
Total	75.92 (6.55)	79.28 (6.67)	77.25 (7.19)	80.89 (10.06)	80.83 (7.33)	85.52 (4.79)		

HA - Hearing aid, CI - Cochlear implant, NH - Normal hearing, SD - Standard deviation

Student's t-test indicated that children with HA had a lower total HRQoL than NH children (p=0.012). Also, every day (school) functioning (p=0.000), family (p=0.013), and friends (p=0.044) subscales were significantly lower in HA compared to NH children (Table 3).

HRQoL of children with CI and NH

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	N	H vs. H	. HA children NH vs. CI child			I childrei	en HA vs. CI children					
	Mean differ- ence	Stan- dard Error	t value	р	Mean differ- ence	Stan- dard Error	t value	р	Mean differ- ence	Stan- dard Error	t value	р
Physical well-be- ing	0.92	3.53	0.26	0.796	-1.69	3.62	-0.47	0.643	-2.61	3.56	-0.73	0.468
Emo- tional well-be- ing	-1.33	3.56	-0.37	0.710	1.86	3.86	0.48	0.632	3.19	3.93	0.81	0.420
Self-es- teem	2.13	3.01	0.71	0.483	2.27	3.28	0.69	0.492	0.14	2.81	0.05	0.959
Family	7.96	3.11	2.56	0.013	9.55	3.26	2.93	0.005	1.59	3.99	0.39	0.692
Friends	13.50	2.66	5.08	0.000	5.00	2.60	1.92	0.061	-8.50	3.10	-2.74	0.009
Every day (School) Func- tioning	6.33	3.06	2.07	0.044	4.52	3.35	1.35	0.184	-1.81	3.26	-0.56	0.580
Total	4.92	1.89	2.59	0.012	3.58	2.13	1.68	0.099	-1.33	2.08	-0.64	0.525

HA- Hearing aid, CI- Cochlear implant, NH- Normal hearing

The mean of the total HRQoL score for the children with CI was 77.25 ± 7.19 , whereas the mean score for the NH children was 80.83 ± 7.33 (Table 2). There was no significant difference when comparing the total scores using an independent samples t-test (p=0.099) (Table 3). When the children with CI and NH were compared, the nearly significant difference between the groups was noted only for the family (p=0.005) subscale.

HRQoL of children with HA and CI

The HRQoL total and sub-scales scores on the KINDL questionnaire of the children using HA and CI were compared, also. Children using HA scored significantly lower only on the friend subscale (p=0.009) (Table 3).

Parents' comparisons in HRQoL sub-scales

Parents of the NH children scored higher in all sub-scales than the parents of children using HA or CI. In the group of HA children, in the opinion of parents, the highest and the lowest rated sub-scales were family contacts and everyday functioning, respectively (Table 2). According to the parents, NH children had better HRQoL than HA children in the total mean (p=0.012), as well as in physical well-being (p=0.000), emotional well-being (p=0.001), self-esteem (p=0.006), and friends subscales (p=0.01) (Table 4).

In CI children, parents awarded the highest points to the Family and the lowest to self-esteem (Table 2). According to the parents, children with CI had a lower HRQoL (p=0.027) and poorer physical (p=0.005) and emotional (p=0.004) well-being compared to NH children (Table 4).When the HA and CI groups were compared, no significant differences were found for any sub-scale or the total score (Table 4).

Table 4.	Comparisons	between parents	s of children w	rith HA, Cl	I or NH in	KINDL-Rsubscales

	Parents of NH vs. HA children		Parents of NH vs. CI children				Parents of HA vs. CI children					
	Mean differ- ence	Stan- dard Error	t value	р	Mean differ- ence	Stan- dard Error	t value	р	Mean differ- ence	Stan- dard Error	t value	р
Physical well-be- ing	10.28	2.46	4.18	0.000	7.99	2.75	2.90	0.005	-2.29	3.11	-0.73	0.465
Emotional well-be- ing	8.88	2.57	3.46	0.001	8.89	2.96	3.01	0.004	0.01	3.35	0.01	0.997
Self-es- teem	6.19	2.15	2.83	0.006	3.97	2.55	1.56	0.125	-2.23	2.62	-0.85	0.398
Family	1.68	2.12	0.79	0.431	0.07	2.72	0.03	0.980	-1.61	2.76	-0.58	0.563
Friends	5.44	2.09	2.61	0.011	3.76	2.64	1.43	0.160	-1.68	2.58	-0.65	0.517
Every day (School) Function- ing	4.97	2.59	1.91	0.060	3.09	3.19	0.97	0.339	-1.89	3.15	-0.59	0.552
Total	6.24	1.43	4.36	0.000	4.63	2.03	2.27	0.027	-1.16	2.09	-0.77	0.443

HA- Hearing aid, CI- Cochlear implant, NH- Normal hearing

Comparison of parent-to-child reports

Spearman's rho was used to assess for correlations between parent and child KINDL questionnaire scores (Table 5). **Table 5.** Spearman's correlation coefficients for the associations between child self-reported and parent-reported HRQoL values

	HA		CI		NH		
	Spearman's rho	р	Spearman's rho	р	Spearman's rho	р	
Physical well- being	-0.182	0.153	-0.160	0.293	0.186	0.152	
Emotional well-being	-0.058	0.653	0.061	0.689	0.296	0.020	
Self-esteem	0.112	0.382	0.196	0.196	0.279	0.030	
Family	0.281	0.026	0.400	0.006	0.017	0.895	
Friends	0.376	0.002	0.110	0.473	-0.002	0.988	

	HA		CI		NH		
	Spearman's rho	р	Spearman's rho	р	Spearman's rho	р	
Every day (School) Functioning	0.344	0.006	0.324	0.030	0.293	0.022	
Total	0.218	0.086	0.222	0.142	0.320	0.012	

For the HA group, significant positive correlations were observed between parent and child responses for the family (r= 0.281), friends (r= 0.376), and everyday functioning (r= 0.344) subscales. For the CI group, Spearman's rho analyses showed significant positive correlations between parent and child responses for the family (r= 0.400) and everyday functioning (r= 0.324) sub-scales. For the NH group, significant positive correlations were observed between parent and child-reported scores for emotional functioning (r = 0.296), self-esteem (r= 0.279), and everyday functioning (r= 0.293) subscales, as well as the total score (r= 0.32). Children using HA or CI provided higher mean scores than their parents, and NH children provided lower mean scores for this sub-scale.

DISCUSSION

Measuring HRQoL in children is complicated for several reasons, and there is wide variability in results obtained in children with HI. For this reason, it's recommended that previously validated instruments be used. We compared the HRQoL of HI children and adolescents fitted with HA or CI with that of age- and sex-matched NH peers using KINDL-R questionnaires. In our study, the overall HRQoL score (75.92) of children with HA was significantly lower than that of NH children (80.83). Children with CI responded similarly, whereas the mean HRQoL score was 77.25. When the children with CI and NH were compared, the difference was nearly statistically significant (p=0.099) and on the family subscale (p=0.005). Overall HRQoL didn't differ between the HA and CI groups in the present study. Several studies have shown that hearing impaired children have a worse quality of life than their healthy peers. Our finding is consistent with the metaanalysis by Roland et al, who found that only school and social categories showed statistically and clinically significant differences between HI and NH children [17]. Niemensivu et al. also concluded that the HRQoL of children with HI was significantly lower than that of NH children [14]. Huber et al. demonstrated that CI children had a lower HRQoL score than NH children at 8-12 years of age [6]. Looi et al. obtained similar results for 4-17-year-old children with CI [7]. Alegre-de la Rosa and Villar-Angulo, using the KINDL-R, found

that HRQoL was significantly better in children with CI than in children with HA [18].

In contrast, other studies have reported that children with HI have similar quality of life to peers with NH. Perez-Mora et al. reported no significant difference in HRQoL between two groups of 58 hearing-impaired children and one group of 30 NH children [19]. Hintermair compared the HRQoL of hearing-impaired students with that of their NH peers and found no significant difference between them [20]. Warner-Czyz et al. found that the QoL of children with CI aged 4 to 7 years was similar to that of NH children [21]. Lovett et al. demonstrated that children with CI aged 1 to 16 years had a similar QoL to NH children [12].

It has been reported that children who use HA or CI, may experience problems in social relationships, particularly with their friends, self-esteem and school sub-scale compared to NH peers. Examination of the individual subscales in the present study revealed that the children with HA reported significantly worse HRQoL than the NH group in everyday (school) functioning, family, and friends subscales. Only the friend subscale score was lower in the children with HA than in the children with CI. Huber and Kipman found that children with HI had significantly more problems with their peers than NH children²¹. Similar findings were reported by Huber [6] and Loy et al. [22] who found lower mean scores on the school, friends and self-esteem subscales for children with CI. Borton emphasized that children with HI may have attention and memory problems and lower school achievement, but this should be monitored individually [23].

In this study, more than 66% of the participating parents were mothers, which is consistent with the literature. Parents of children with NH scored better on all subscales than parents of children with HI. Parents estimated that children with NH had significantly better HRQoL in the total mean and physical wellbeing, emotional well-being, self-esteem and friends subscales than children with HA. In addition, children with CI had lower HRQoL and poorer physical and emotional well-being than NH children. According to the literature, parents of healthy children tend to overestimate their children's QoL, while parents of children with chronic illness underestimate their QoL. Zhumabayev et al. [10] and Kumar et al. [24] reported that parents of children with CI assessed HRQoL positively in all subscales.

We found statistically significant positive correlations between parents' and children's responses on the family, friends and everyday functioning subscales for the HA group. For the CI group, significant positive correlations were found between the parent and child reported scores for the family and everyday functioning subscales. Significant positive correlations were found in the NH group for the total scores and the subscales emotional functioning, self-esteem and everyday functioning. Our result is consistent with the results of Eiser and Varni [25].

The limitations of our study were the relatively small sample size and the unequal number of participants in each group. We could not stratify participants by age, speech development or auditory measures. Another limitation was that parents could not be matched on socio-demographic characteristics. On the other hand, our study was the first assessment of HRQoL of children with HI in Montenegro. We emphasize the importance of children's assessment of HRQoL rather than relying only on parents' opinions. Strengths of this study include the use of a generic HRQoL instrument that is used worldwide.

CONCLUSION

Using a generic HRQoL questionnaire, our results showed that the children with HA generally had significantly lower child and parent- HRQoL scores than NH children. We found that children with CI can achieve similar HRQoL to NH peers, and that children with HA assess HRQoL similarly to children with CI.

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