Parents' perception of Health-Related Quality of Life in Children with Cochlear Implants: The Impact of Language Skills and Hearing

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Abstract

Purpose

The study compared how parents of children with cochlear implants (CI) and parents of children with normal hearing perceive their children's Health-Related Quality of Life (HR-QOL).

Method

The sample consisted of 186 Norwegian speaking children in the age span 5;0 to 12;11: 106 children with CIs (53% boys) and 80 children with normal hearing (NH) (44% boys). No children had known additional disabilities affecting language, cognitive development or HR-QOL. Parents completed the generic questionnaire Pediatric Quality of Life Inventory™ (PedsQL™), while children completed a test battery measuring different aspects of language and hearing.

Results

Parents of children with CIs reported statistically significantly poorer HR-QOL in their children, on PedsQL Total score and the subdomains Social functioning and School functioning. Roughly 50% of parents of children with CIs reported HR-QOL levels (Total score) within normal limits. No significant differences between groups emerged on the Physical health and Emotional functioning subscales. For the children in the CI group, better speech perception in everyday situations was associated with higher proxy-ratings of HR-QOL. Better spoken language skills were weakly to moderately associated with higher HR-QOL.

Conclusion

The findings suggest that the social and school situation is not yet resolved satisfactorily for children with CIs. Habilitation focusing on spoken language skills and better sound environment may improve social interactions with peers and overall school functioning.

Introduction

Hearing loss is a disability that can have broad-ranging effects on the everyday functioning of children and their families. One of its most paramount effects is language difficulties, caused by the reduced ability to acquire and communicate effectively using spoken language (Hoffmeister & Caldwell-Harris, 2014). Previous studies, across different countries, indicate the same tendency: many deaf students without CIs leave school with poor reading abilities (Conrad, 1977), are less likely to go on to higher education (Blanchfield, Feldman, Dunbar, & Gardner, 2001; Eide & Gundersen, 2004; Woodcock & Pole, 2008), have a lower labor participation as adults (Blanchfield et al., 2001; Eide & Gundersen, 2004; Woodcock & Pole, 2008), and a higher frequency of mental health problems compared to the normal hearing population (Fellinger et al., 2005; Kvam, Loeb, & Tambs, 2007).

In recent years, *cochlear implants* (CIs) has become an increasingly common aid for children born with sensorineural deafness in the western world. A cochlear implant is a hearing device that, through a surgically inserted electrode array in in the cochlea, provides the acoustic nerve with electronic stimulation that is perceived as sound by the auditory cortex (Stach, 2010). CIs has enabled many children, and adults, to take part in mainstream society and education to a greater extent than what was previously possible for deaf individuals, by providing them the ability to hear and acquire spoken language (Duarte, Santos, Rego, & Nunes, 2016; Huber, Wolfgang, & Klaus, 2008; Spencer, Tomblin, & Grantz, 2012). While it is well established that cochlear implantation represents a substantial medical advance, and many children who received CIs early in life can acquire age-appropriate language skills (Fulcher, Purcell, Baker, & Munro, 2012; J. Leigh, Dettman, Dowell, & Briggs, 2013), less is known about how these advances affect their everyday functioning and quality of life (Lin & Niparko, 2006; Morettin et al., 2013). Health-Related Quality of Life (HR-QOL) is a multi-dimensional concept, commonly used within research to measure an individual's subjective perception of

physical and mental health, and social well-being. The concept may also include measures of the individual's relation to and perception of salient features in their environment, such as school or work (Berzon, Hays, & Shumaker, 1993; The WHOQOL Group, 1995; World Health Organization, 1948). Though a number of studies have been published on HR-QOL in children with CIs during the last decade, the overall empirical picture with regards to results is still mixed.

HR-QOL in Children with CIs compared to Children with Normal Hearing

Some studies that look into HR-QOL in children with hearing loss find similar levels of HR-QOL as is found in normal hearing peers see e.g. (Domellof, Hedlund, & Odman, 2014; Duarte, Santos, Rego, & Nunes, 2014; Hintermair, 2011; Kumar, Warner-Czyz, Silver, Loy, & Tobey, 2015; Lovett, Kitterick, Hewitt, & Summerfield, 2010; Loy, Warner-Czyz, Tong, Tobey, & Roland, 2010; Meserole et al., 2014; Perez-Mora et al., 2012; Warner-Czyz, Loy, Roland, Tong, & Tobey, 2009). Other studies present results that are not definite or indicate reduced levels of HR-QOL in children with CIs in one or more domains, particularly with regards to psychosocial well-being, school well-being or peer-problems see e.g. (Fellinger, Holzinger, Sattel, & Laucht, 2008; Hintermair, 2011; Huber, 2005; Petrou et al., 2007; Rachakonda et al., 2014; Razafimahefa-Raoelina et al., 2016; Schick et al., 2013; Wake, Hughes, Poulakis, & Collins, 2004; Wake, Hughes, Poulakis, Collins, & Rickards, 2004).

In a recent review of the literature by Roland et al. (2016) the authors note that the large variability in HR-QOL instruments make it difficult to draw conclusions regarding children with hearing loss. In the four studies that used the generic questionnaire PedsQL the authors were able to perform a meta-analysis. Results showed lower levels of HR-QOL in domains measuring social and school functioning in children with hearing loss compared with normal hearing peers.

Which Factors may be Associated with HR-QOL in Children with Hearing Loss?

Nowadays most children with a hearing loss in Norway and other western countries are integrated in regular schools in their community. Good speech perception and spoken language skills will therefore be essential to communicate effectively. Better communication skills and the use of oral communication have been found to be associated with a reduced incidence of mental health problems and reduced peer problems (Percy-Smith et al., 2008; Theunissen et al., 2011; Theunissen et al., 2015). Similarly, in a study by Fellinger, Holzinger, Beitel, Laucht & Goldberg (2009) children who had better spoken language skills had significantly fewer problems with peer relations in mainstream schools, but more peer problems in segregated schools for the deaf. The reverse was found for children proficient in sign language, thus indicating that efficient communication in the same mode as the other children at school is important for development of peer relations (Fellinger, Holzinger, Beitel, et al., 2009). Thus, it is reasonable to assume some form of relationship between HR-QOL and language skills in children with CIs (Morettin et al., 2013).

Furthermore, some children with CIs have relatively poor speech perception, not only because of their hearing loss, but also due to factors such as less well developed cognitive skills (e.g. vocabulary (Lund, 2015) or working memory (Lyxell et al., 2008)) and is especially obvious in noisy situations. This is challenging for children, as much learning, both language acquisition in general as well as academically, often takes place in noisy environments such as schools, playgrounds or in conversations with more than one person. Language difficulties and problems with speech perception may affect both psychosocial functioning (Huber & Kipman, 2011; G. Leigh et al., 2015) and physical health, e.g. through headaches due to exhausting listening conditions or shoulder and neck pain (Anmyr, Olsson, Larson, & Freijd, 2011; Sæbø, Wie, & Wold, 2016).

Children diagnosed and implanted at an early age have increased chances of developing spoken language abilities at an age appropriate level (Manrique, Cervera-Paz, Huarte, &

Molina, 2004; Tobey et al., 2013; Wie, 2010), and may thus have different levels of HR-QOL than later implanted children with poorer language skills. Korver et al (2010) investigated the effect of early new-born hearing screening on general development, spoken language and HR-QOL. The results showed that the earlier identified children had a larger spoken vocabulary, better scores on social development and significantly higher HR-QOL than the later identified group (Korver et al., 2010). Similar results have been found in studies where lower age at first amplification (Loy et al., 2010; Schorr, Roth, & Fox, 2009) was related to higher levels of HR-QOL in children with CIs. However, neither Perez-Mora et al. (2012) nor Kumar et al. (2015) found correlations between HRQOL and background variables such as chronological age, age at CI activation/audiological treatment or duration of device use/CI use.

Another factor found to influence spoken language abilities and speech perception skills in children with hearing loss is nonverbal IQ (Geers, 2002; Wie, Falkenberg, Tvete, & Tomblin, 2007). It is critical to obtain measures of nonverbal abilities in children with hearing loss, as it is estimated that 30 to 40% of these children have additional disabilities, such as intellectual disability, autism spectrum disorders, or syndromes affecting development in several areas that might also affect development of non-verbal IQ (Birman, Elliott, & Gibson, 2012; Stacey, Fortnum, Barton, & Summerfield, 2006). Presence of additional disabilities has also been found to increase the risk for experiencing reduced levels of HR-QOL, or related concepts such as problems with peer interaction, compared to children with hearing loss without additional disabilities (G. Leigh et al., 2015; Necula, Cosgarea, & Necula, 2013; Rajendran & Roy, 2010; Zaidman-Zait, Curle, Jamieson, Chia, & Kozak, 2017). The presence of additional disabilities is thus a known factor associated with reduced HR-QOL.

Higher socioeconomic status, commonly measured through maternal education, is often associated with better spoken language abilities (Szagun & Stumper, 2012). As with other variables affecting language abilities, it could be expected that this variable would be

associated with higher levels of HR-QOL. Higher levels of maternal education may also be associated with better access to health care and economical resources. Some studies do find higher SES to be associated with higher levels of HR- QOL or related concepts in children with hearing loss (Kirman & Sari, 2013; Sahli, Arslan, & Belgin, 2009), whereas other studies do not find this association (Huber & Kipman, 2011; G. Leigh et al., 2015; Necula et al., 2013).

Why are previous findings inconsistent?

A reason why previous research is inconsistent with regard to differences in HR-QOL between children with hearing loss and typically developing peers, may be that the groups examined in these studies vary according to a number of factors, such as age, type of amplification and mode of communication. Additionally, there is also large heterogeneity *within* each study sample, including children with hearing losses of different magnitude and age of occurrence, unilateral and bilateral CIs, use of different technology such as hearing aids or CI or a combination e.g. (Fellinger et al., 2008; Hintermair, 2011; Meserole et al., 2014; Perez-Mora et al., 2012; Petrou et al., 2007; Wake, Hughes, Poulakis, & Collins, 2004). Heterogeneity amongst the sample of children participating in a study can complicate and affect the possibility to draw conclusions for a number of reasons. For example, bilateral implantation has been found to be advantageous compared to unilateral implantation with regards to spatial hearing, speech perception in noise and language abilities (Boons et al., 2012; Lovett et al., 2010). Moreover, the degree of hearing loss influences language development (Tomblin et al., 2015). Some studies also indicate that use of CIs is associated with higher levels of HR-QOL or lower frequency of mental health problems compared to children using hearing aids, with one possible reason being the better and more extensive habilitation these children often receive (Necula et al., 2013; Theunissen et al., 2015). Variation with regards to factors such as age at amplification, communication mode (sign vs. spoken language), presence of additional disabilities and large chronological age span may also affect the outcome. Some studies

investigate small samples of 20-30 children, and they may thus not have sufficient statistical power to detect even medium effect sizes for the comparison with normative data or control groups.

Heterogeneity in study design is another aspect that may contribute to explaining the variability in previous findings. Some studies have relied on norming data rather than including a control group. Collecting data from both the clinical and the typically developing group in the same study allows for a greater control over factors that can influence results, e.g. variables associated with data collection such as instructions to participants. Moreover, a control group enables the comparison of background variables that are not part of the norming study, but that may influence quality of life, such as cognitive or linguistic functioning.

Which questionnaires the different studies use can also affect results. When investigating HR-QOL in a clinical population, there are two major approaches to the assessment of HR-QOL: *disease specific* or *generic* questionnaires. Disease specific questionnaires focus mainly on issues considered to be frequently experienced in a clinical population. Generic questionnaires investigate domains of HR-QOL thought to be important regardless of illness or disease (Fayers & Machin, 2007). In line with the definition provided by the WHO, generic questionnaires should at least include domains measuring physical-, emotional- and social well-being. In several of the studies, domains such as school functioning or family well-being are also included, as these domains are often considered salient in the children's environment (Berzon et al., 1993; The WHOQOL Group, 1995; World Health Organization, 1948). The differences between studies in which specific domains of HR-QOL they include, is therefore one factor that may explain variations in outcomes between studies. It may also be the case that different combinations of domains in generic questionnaires measure the essence of HR-QOL more or less well. Nevertheless, standardized generic questionnaires do offer the possibility to compare HR-QOL reports of a clinical population to that of the

general population. This is important, as studies has revealed that individuals who are deaf or hard of hearing have more frequent mental health issues such as anxiety or depression compared to the population as a whole (Kvam et al., 2007; Theunissen et al., 2011), and such conditions likely affect HR-QOL in children (Fellinger et al., 2008). Studies using generic questionnaires can thus provide valuable information on which areas interventions need to target and where better help-services are needed.

Another difference in how studies were conducted is the use of self- vs. proxy-reports. In studies of HR-QOL, self-report is recommended in many cases. While proxy-reports cannot serve as a direct substitute for self-reports, proxy-reports can provide valuable insight and perspective on HR-QOL in very young children, children with language impairments or in clinical groups that are unable to self-report for different reasons (Matza, Swensen, Flood, Secnik, & Leidy, 2004; Varni, Limbers, & Burwinkle, 2007). Even in children with CIs who have normal nonverbal abilities, language difficulties are common (Löfkvist, Almkvist, Lyxell, & Tallberg, 2014; van Wieringen & Wouters, 2015). Parents of children who have chronic health issues, have been found have more insight and to be more reliable reporters on HR-QOL than parents of typically developing children (Eiser & Jenney, 2007).

It is thus important to that studies of HR-QOL in children with hearing loss are transparent on how data was collected and which children were included in the study. Investigating factors beyond HR-QOL, such as how language abilities, speech perception, age of implantation or SES affects HR-QOL, may also contribute to our understanding of why some children experience enhanced or diminished HR-QOL. In the present study, we therefore aim to narrow down the heterogeneity by using precise and strict inclusion criteria.

The Purpose of this Study

The aim of this study was to investigate generic proxy-reported HR-QOL in a large sample of children with CIs. As shown in the literature review, there is a need for larger studies that

compare HR-QOL in well-described samples of children with CIs to control groups of normal hearing peers, investigating factors beyond HR-QOL. Thus, the current study investigated a relatively homogenous group of children with few other known factors affecting HR-QOL than the presence of their hearing loss. This enabled us to focus specifically on the impact of hearing loss on HR-QOL. In addition, we used a well-described questionnaire previously used to measure HR-QOL in clinical groups of children, including children with hearing loss. The study addressed the following two research questions:

1. How do parents of children with CIs perceive the HR-QOL of their children, compared to parents of children with normal hearing and typical development?
2. To what degree can variation in proxy-reported HR-QOL in children with CIs be explained by the children's language skills, nonverbal IQ, speech perception or background variables such as communication mode, socioeconomic status and age at implantation?

Materials and Methods

The study had a cross-sectional design and was part of a larger national study: *Speech perception, language and quality of life in people who have received CI as children in Norway*. The project was approved by the Regional Committees for Medical and Health Research Ethics in Norway and the Data Protection Official at Oslo University Hospital. The project extended an invitation to the entire Norwegian population of CI users implanted before the age of 18 in Norway during the period 1988 to 2015. This larger study had 496 participants, out of a possible sample of 606. Thus, the final participation rate was 82%.

Sample

The inclusion criteria used in the present study were set to ensure that the group of children with CIs had few known factors other than hearing loss affecting HR-QOL and had similar

characteristics with regards to language background, bilateral stimulation and relatively early implantation.

All participants, in both the CI and NH groups, met the following three inclusion criteria: 1) All had a standard score of 75 or above on the Raven's Progressive matrices (Raven, 2004, 2008) (a test of nonverbal IQ). This criterion was set to prevent including children who have intellectual disabilities (defined in DSM V as IQ scores below 70, including a margin of measurement error; (American Association on Intellectual and Developmental Disabilities; American Psychiatric Association, 2013). 2) The children had no known additional disabilities known to affect language development or HR-QOL. Children who had additional cognitive, physical and communicative disabilities possibly affecting different aspects related to quality of life, such as Autism spectrum disorder, ADHD, cerebral palsy, diabetes, epilepsy, cancer or chronic illnesses, were excluded. 3) The child, and at least one of the child's parents, should have spoken Norwegian as their native language.

Out of the 206 children with CI in the age range 5 to 12 years old who agreed to participate in the national project, there were 106 who fulfilled the inclusion criteria for the present study, which is approximately half of the children in the relevant age span in the national study. The main reasons for exclusion were nonverbal IQ below 75 (n= 44), the presence of an additional disability thought to affect cognitive or language development or HR-QOL (n= 32), and other first language than Norwegian (n= 24).

The final sample consisted of 186 children in the age span 5;0 to 12;11 years:106 children with CI (56 boys, 53%) and 80 children with normal hearing (NH) (35 boys, 44%). A power analysis showed that this sample provided adequate statistical power (.8) to detect a medium-sized effect of .35 (G*Power software; Faul, Erdfelder, Buchner, & Lang, 2009). An additional inclusion criterion for the NH group was the presence of *otoacoustic emissions* indicating normal inner ear function and normal hearing with hearing thresholds better than 30

dB HL. The CI and NH groups did not differ significantly with regards to chronological age. There was a statistically significant group difference in nonverbal IQ, though a difference of this magnitude is not regarded as a clinically relevant (see Table 1).

Children in the CI group had somewhat lower levels of *socio economic status* (SES) than children in the NH group, as measured by the parents' level of education. Sixty-three percent of the mothers in the CI group had completed at least one year of university or college, compared to 84% of the mothers in the normal hearing group.

Characteristics of the children in the CI group. The largest subgroup in the sample (55% of participants) consisted of children who were prelingually deaf, i.e. they were either born deaf or became deaf within 12 months after birth. Children who became deaf between 13 and 35 months accounted for 8%, and 25% were born with a hearing loss that progressed into profound deafness and thus required treatment with CIs. For 12% of the children onset of hearing loss is unknown. Most children with an unknown onset of hearing loss were born before the universal hearing screening was implemented in Norway.

The majority of the children were implanted at an early age. There were 19 children who were implanted before 12 moths. Amongst the children who were prelingually deaf, 68% were implanted by age 2 years, and 90% had received their implants by age 3 years. Amongst the 36 children who were implanted after the age 3, only five were prelingually deaf, thus the remaining children either had normal hearing or some residual hearing at age 1 year. The youngest children that have been implanted in Norway were five months old. It is an explicit goal that children who are diagnosed early should be implanted in time to give them access to sound before they are 12 months old.

Sixty-eight percent of the parents reported that they mainly used spoken language when communicating with their children, whereas 17% reported that they occasionally used some sign support while speaking, supporting the main words in the sentences. The remaining

parents reported that they either used spoken language with a higher level of sign support (4%), both sign language and spoken language (1%), mainly sign language (1%) or used a combination of different language modes (9%). All children were communicating through spoken language with or without sign support at some degree, including the child who predominately communicated with the parents through sign language.

None of the children in the sample had unilateral auditory stimulation. In Norway, the government covers the cost of implantation, and bilateral implantation is offered as the standard procedure. Thus, bilateral cochlear implants were worn by 86% of the children, of which 67% received simultaneous bilateral implants and the remaining were sequentially implanted. Fourteen percent of the children in the CI group used a cochlear implant in combination with a hearing aid.

The cause of deafness was identified for 62% of participants with CI, with Connexin 26 being the most common etiology present in 19% of the participants. Other more common causes of deafness were Pendred or LVAS (*Large Vestibular Aqueduct Syndrome*) (10%) and Meningitis (9%).

The majority of parents in the study (75%), reported that they had received individual guidance on how they could support their children's language development after their children received the implant. The amount of guidance they had received varied from once a week (15%), once a month (29%), to approximately five times a year (31%). Note, however, that 23% of parents reported that they had never or seldom received any guidance after implantation, and 2% did not report how much support they have received.

Most (87%) of children in the study received some form of additional educational support in school, while the remaining 13% reported that they did not receive any additional support in school or kindergarten. According to parent-reports, 38% of the children who received support had less than five hours a week of special needs education, while 39%

received more than five hours a week. Eighty-six percent of the children attended mainstream school, while the remaining 14% were educated in schools for the deaf.

Recruitment and Procedure

The children with CIs were recruited at the National CI Centre at Oslo University Hospital, Norway. Children in the normal hearing group were recruited from different schools in the south-eastern part of Norway, from both urban and rural areas. The children in the CI group were tested in conjunction with their annual appointment at the cochlear implant center, and their parents completed the questionnaires at the clinic while waiting for their children to complete their tasks. Due to this coordination of appointments, the time spent completing the assignments in the research project varied somewhat between children. Thus, not all tests were completed by the children at the same time as the HR-QOL questionnaire completed by the parents in the CI group. For 16 children, there was a discrepancy of more than 6 months between testing with the British Picture Vocabulary Test-II and proxy-report. For six children, there was a discrepancy of more than 6 months between the HR-QOL questionnaire and testing with the Clinical Evaluation of Language Fundamentals-4. To check if this issue affected reliability and validity of the results, all analysis on language data and HR-QOL were also conducted excluding these participants. Since there was no significant difference in the results, all results presented in this article thus include these participants.

The children in the normal hearing group were primarily tested at their schools, though some were tested at the cochlear implant center or at home.

Materials

All children completed a battery of tests assessing hearing, language abilities and cognition. HR-QOL and demographic variables were assessed through questionnaires. Medical records

from the hospital provided information on date of operation(s), time of hearing loss as well as hearing loss etiology.

Health-related quality of life. HR-QOL was assessed trough the Norwegian translation of the Pediatric Quality of Life Inventory™ 4.0 (PedsQL™) generic core scale (Varni et al., 2007). The questionnaire exists in several age specific versions, which enables the questionnaires to be used in different age groups. The Norwegian version of the questionnaire is validated for use in adolescents between 13 and 15 years of age, where it was found to be a valid and reliable (Reinfjell, Diseth, Veenstra, & Vikan, 2006). There are thus no national norming data available for the age group examined in the current study. We compensated for the lack of norming data by collecting data from a group of 80 normal hearing children.

In the current study a Norwegian translation of the questionnaire's proxy-report version for younger children (5-7 years) and children (8- 12 years) was used. Like the original English version of the questionnaire, it consists of 23 questions, which assess four domains of HR-QOL: Physical functioning (8 items), Emotional functioning (5 items), Social functioning (5 items), and School functioning (5 items). In addition, the questionnaire provides two composite scores: a Total score including all items of the questionnaire and a Psychosocial health score (15 items) derived from a combination of the scores of the subscales Emotional, Social and School functioning. The parents rate how they perceive their children's HR-QOL by evaluating how much of a problem different items have been for the past month on a Likert scale ranging from never (0), almost never (1), sometimes (2), often (3) or almost always (4). Upon scoring, the scale is reversed and linearly transformed to a 0 to 100 scale (0=100, 1=75, 2=50, 3=25, 4=0). A higher score indicates higher levels of HR-QOL.

PedsQL 4.0 is designed with respect to the core definition of health and quality of life provided by WHO. In addition to the minimum concepts physical, mental and social health, the questionnaire includes school functioning, which is considered a salient feature in children's

everyday life according to the WHO definition of QOL (Varni, Seid, & Kurtin, 2001; World Health Organization, 1948).

Scale reliability. Cronbach's alpha was calculated to assess reliability of the questionnaire, as well as reliability for each of the subscales. A coefficient greater than 0.7 was considered to be satisfactory (Field, 2013). Cronbach´s alpha was found to be larger than .70 for both the questionnaire as a whole, as well as for each subscale (see Table 2). Table 2 shows Cronbach's alpha for the group of participants as a whole, but there were similar levels in each of the groups separately. In the normal hearing group, the overall alpha was .891, while it was .897 in the CI group.

The PedsQL has been designed to measure four different subscales as well as a composite score and a total score including all questions. Though the subscales measure different factors, they also measure the larger multidimensional construct HR-QOL. Some intercorrelations amongst the subscales would therefore be expected. The observed intercorrelations (see Table 2) were found to be similar to those reported in the Norwegian validation of the PedsQL questionnaire for adolescents (Reinfjell et al., 2006).

Language and cognitive measures. Nonverbal IQ was assessed through the Raven's Colored Progressive Matrices (Raven, 2004) for the younger children, and through the Raven's Progressive Matrices Educational Plus (Raven, 2008), for children older than 9:0 years. The test gives a measure of nonverbal IQ, and as the test has simple instructions and all stimulus materials are nonverbal, it is often used in groups with language and communication impairments.

The Norwegian version of the British Picture Vocabulary Scale 2 (BPVS-2) was used as a measure of receptive vocabulary (Dunn, Dunn, Whetton, & Berley, 1997; Lyster, Horn, & Rygvold, 2010). In this test, the children are provided a single spoken word, and asked to

choose the one picture amongst four different pictures that most closely corresponds to the target word.

In order to obtain a measure of broader language skills, the children completed the Norwegian version of the Clinical Evaluation of Language Fundamentals 4 (CELF-4) (Semel, Wiig, & Secord, 2003). This is a comprehensive test that provides a range of different language measures and which has been validated and normed for Norwegian. The Norwegian version of the test incudes 13 subtests that can be combined into seven separate composite indexes, measuring different aspects of receptive and expressive language such as semantics, grammar, syntax, phonological memory and working memory. The test was administered in full version to all children who participated in the project.

Hearing. The speech perception abilities of the children in the CI group were measured by means of a monosyllable repetition test, *the Phonetically Balanced Word Lists* (PB-N) (Øygarden, 2009) and the Norwegian adaptation of *the Hearing in Noise Test* (HINT) for children (Myhrum, Tvete, Heldahl, Moen, & Soli, 2016). In PB-N test the participant is asked to repeat 50 single syllable words, and a score is estimated as the percentage of correct responses. In the HINT, the participant is asked to repeat sentences in two test conditions: first in quiet and then in background noise. In the quiet condition the sound was constant at 65 dB, and the score was the percentage of words in sentences repeated correctly. The sound to noise ratio score (S/N) was calculated with an adaptive test procedure, indicating the threshold where the participant could repeat 50% of sentences correctly in a noisy environment (Myhrum et al., 2016). Only participants who had a score of more than 75% in the quiet condition, were tested in the noise condition. There were 39 children who scored below 75%, and thus only the remaining 67 children were included in the analysis involving the hearing in noise data. All hearing tests were completed in an anechoic chamber under standardized testing conditions.

The parental questionnaire on background variables also included seven questions on

functional hearing adapted for the study from the Speech, Spatial and Qualities of Hearing Scale (SSQ) (Gatehouse & Noble, 2004). The parents indicated if the child could hear or perceive speech/sound in different everyday situations on a Likert-scale ranging from 0 (not able to) to 10 (without problems) e.g. *The child is talking with one other person in a quiet, carpeted lounge-room. Can he or she follow what the other person says?* (Gatehouse & Noble, 2004, p. 96). The questions from the SSQ were combined to one index score for the analyses. This index had a Cronbach's alpha of .861.

Statistical Analysis

Means, medians and standard deviations or IQR were calculated for all variables. The data violated assumptions of a normal distribution. This is common in studies of HR-QOL, for two reasons. First, this measure typically does not assume equal distribution over the different response categories. Second, some of the items often take extreme values. Depending on the concept that is measured, one will often anticipate scores at the higher or lower end of the scale (Fayers & Machin, 2007). Appropriate nonparametric statistical tests were therefore applied: the Mann-Whitney test to compare scores across groups, and Spearman's correlations to explore associations between variables. An alpha level of .05 was chosen to determine statistical significance.

Results

Comparing HR-QOL Scores in the NH and CI group

Analyses revealed significantly lower scores in the CI group than in the NH group on the PedsQL subscales Total score, Social functioning, School functioning and Psychosocial health. The parents of children with CIs, thus, perceived them as having poorer HR-QOL than parents of normal hearing children in several domains. No significant difference between groups emerged on the subscales Physical health and Emotional functioning (see Table 3 for results).

See Figure 1 for group median comparison and group score distribution for each of the PedsQL subscales and Total score.

In previous studies using the PedsQL, "at-risk" status for impaired HR-QOL has been proposed to be 1 SD below the mean of the total population, and the minimal clinically important difference for change in the Total score has been suggested to be 4.5 points (Varni, Burwinkle, Seid, & Skarr, 2003). In the current study, the differences between the groups' means exceeded the minimal clinical important difference on the Total score. At-risk status was estimated as a score more than 1 SD below the NH group's mean score, as there are no national norms for the general population in Norway. For the Total score, the normal hearing group´s mean was 87.91 and the SD was 9.17, thus indicating that a score below 78.74 represented an at-risk score. In the normal hearing group 81% of the children had scores equal to 1 SD below mean or higher. In the CI group, this number was 57%, indicating that just over half of the children in the CI group had an overall HR-QOL that is within normal limits.

For the two domains where children in the CI group were reported to have similar levels of functioning as their normal hearing peers, the majority of children had scores equal to 1 SD below NH groups mean or higher. For the Physical health domain 79% of the children had scores equal to 1 SD below mean or higher, whereas this was 75% in the Emotional functioning domain.

In the two domains where proxy-reports indicated poorer HR-QOL than the control group, half of the children in the CI group were reported to have scores equal to 1 SD below mean of the normal hearing children or higher. In the Social functioning domain and the School domain, 56% and 48% respectively, were reported to have good HR-QOL. On the composite score, Psychosocial health, 48% of the children were reported to have good HR-QOL.

The parents of the normal hearing children in the current study, however, reported very few problems with peer interaction. Forty-four of the 80 participants (55%) in the normal

hearing group reported that their children never experienced any difficulties in social settings, and thus answered all the five questions in the Social functioning domain with "never." In comparison, only 15 (14%) of the parents in the CI group did the same.

What Variables are Associated with HR-QOL in Children with CI?

Additional analyses were carried out to examine the factors or variables affecting HR-QOL outcomes for the CI group (n =106).

Background Variables. In the data analysis, SES was estimated both as a mean of the educational level of the mother and father combined, and each parent separately.

The analysis of different background variables showed no significant correlation between HR-QOL and age at diagnosis of hearing loss, age at implantation, chronological age, nonverbal IQ, communication mode, score on the single syllable word identification test or SES.

Speech perception and functional hearing. There were some weak negative statistically significant correlations between scores on the HINT (a lower score equals lower S/N ratio), and the Total score r= -.275, n=67, p=.024 and School functioning r= -.244, n=66, p=.048 (list one of two completed, unadjusted for age), indicating that children who hear better in noise also had higher scores of HR-QOL in these domains. When age was partialled out in the correlations, they were no longer significant (Total score r= -.230, n=64, p=.063 and School functioning r= -.233, n=63, p=.062) though still indicating a tendency for children who hear better in noise to have a somewhat higher overall HR-QOL.

Parent-report of their child's functional hearing (questions adapted from the SSQ and combined into one index of functional hearing), was correlated with all subscales and composite scores of the PedsQL. The strongest correlation was between the functional hearing index and the Psychosocial Health scale, r= .394, n=105, p<.000. These moderate correlations indicate that better hearing in different real-life situations is associated with higher HR-QOL.

Language measures. There were no significant correlations between the receptive vocabulary measure BPVS and the PedsQL scores (see Table 4). There were, however, several significant correlations of weak to moderate strength between CELF indexes and PedsQL subscales (see Table 4). The strongest correlations were between the CELF core, language memory, and working memory scales, and the PedsQL domains Social and School functioning, as well as the two composite scores Total score and Psychosocial health. There were no significant correlations between language measures and the PedsQL subscales Physical health and Emotional functioning (see Table 4). This indicates that better language and communication abilities were associated with higher proxy-rating of social and academic functioning.

Discussion

This study aimed to investigate generic proxy-reported HR-QOL in Norwegian children with CIs. Precise and strict inclusion criteria were formulated in order to reduce the impact of other factors known to affect HR-QOL. This was an important aim of the present study given that many previous studies of HR-QOL have used heterogeneous samples of children with hearing loss.

Comparing HR-QOL Scores in the CI and NH Groups

On the subscales Physical health and Emotional functioning, parents of children with CIs and parents of children with NH evaluated their children to have similar levels of HR-QOL. However, on the subscales Social functioning and School functioning, parents of the CI group reported significantly lower levels of HR-QOL. Furthermore, both the Total score indicating overall HR-QOL and the Psychosocial health score were significantly lower in the CI group than in the NH group.

The results in the present study are in line with previous findings in the literature that find diminished HR-QOL in children who have a hearing loss, particularly with regards to Psychosocial, Social- or School functioning e.g. (Fellinger et al., 2008; Rachakonda et al., 2014; Roland et al., 2016; Wake, Hughes, Poulakis, & Collins, 2004; Wake, Hughes, Poulakis, Collins, et al., 2004). However, in some of these studies the variation in the results might be explained partly by factors like relatively high mean age at intervention or implantation (Fellinger et al., 2008; Wake, Hughes, Poulakis, Collins, et al., 2004), heterogeneity in the sample including children with additional disabilities or a variation in type of amplification (Rachakonda et al., 2014; Wake, Hughes, Poulakis, & Collins, 2004). In the current study, all of these variables were either controlled for through exclusion criteria, or by applying separate analysis of subgroups. As the children with CIs in the current study differed from their normal hearing peers on few other variables than their hearing loss, one might have expected to find that they differed less on HR-QOL compared to normal hearing peers than in many of the previous studies.

A closer examination of the results in the CI group revealed that more than half (57%) of the children are reported to have good functioning on the questionnaire's Total score, and most of the children are doing well on the Physical health (79%) and Emotional functioning (75%). Emotional functioning is the subdomain which may best reflect different aspects associated with mental health and includes questions on whether the child is feeling sad or worry a lot. Problems associated with mental health issues are found in studies of children who have a hearing loss in general (Fellinger et al., 2008; Van Eldik, 2005). However, studies focusing solely on children using CIs indicate that this population might more closely resemble children with NH (Huber & Kipman, 2011; Theunissen et al., 2012), which is in line with the results of the current study. The finding that most of the children with CIs are reported not to

struggle with Emotional functioning is a positive finding, which might indicate that this aspect of the psychological well-being of this group is not diminished.

It seems, however, that Psychosocial health, Social functioning and School functioning are perceived by the parents as areas where their children are struggling. Approximately half of the children in the CI group are reported to have a Social (56%) and School functioning (48%) comparable to that of their normal hearing peers. It is possible that the parents of the normal hearing group overestimate how well their children are doing, as the score on the Social domain is high compared to both the American norming sample of the PedsQL questionnaire (Varni et al., 2007) and parent-reports from the validation of the questionnaire in a Norwegian sample of typically developing adolescents (Reinfjell et al., 2006). There are also differences in the SES in the two groups that might affect how similar the two groups are, and these differences might be especially relevant for school functioning. The results can nonetheless indicate suboptimal levels of functioning for the majority of children with CI in the domains of social interaction and school functioning. The problems with peer relations that are reported by the parents of the CI group are in line with results reported in previous studies that uses questionnaires that target specifically mental health and social skills in children and adolescents with CIs (De Giacomo et al., 2013; Huber et al., 2015).

Another important factor in the current study is the use of proxy-reports. Though parents of children with chronic health issues have been found to report more reliably than parents of typically developing peers (Eiser & Jenney, 2007), it also seems that proxies tend to overestimate the impact of symptoms on functioning in children with chronic health conditions (Upton, Lawford, & Eiser, 2008). Thus, proxy-reports do not necessarily have a high concordance with self-reports. In some studies of children with CIs, parents have been found to be reliable reporters of HR-QOL (Loy et al., 2010), while others have found that parents tend to underestimate their children's well-being (Warner-Czyz et al., 2009). In the current study we do

not know the children's perspective and should thus keep in mind the possibility of parents both under- or overestimating the children's HR-QOL.

The findings of the current study still indicate that though many children have a good HR-QOL, the children's situation in school and social settings is not yet resolved in a satisfactory way, even in a group of children with few other known difficulties besides their hearing loss.

Which variables predict HR-QOL in children with CI?

The current study did not find any association between HR-QOL, and different background variables previously found to be of importance, such as age at identification or audiological intervention (Korver et al., 2010; Loy et al., 2010; Schorr et al., 2009), spoken language as main communication mode (Percy-Smith et al., 2008; Theunissen et al., 2011; Theunissen et al., 2015), or higher levels of socioeconomic status (Kirman & Sari, 2013; Sahli et al., 2009). There might be several reasons why these variables were not related to HR-QOL in the group of children with CIs investigated in the current study. Within the group of children who are prelingually deaf, 69% were implanted by two years of age, and 91% were implanted by three years of age. This is relatively early compared to many previous studies. Furthermore, only 14% of the children in the CI group attended either a class or school for the deaf, while the majority of children attended mainstream schools and used spoken language as their main mode of communication. None of the children in the study relied solely on sign language, though some were dependent upon some sign support. The lack of variation in the group on these variables might thus explain why they were not strongly associated with HR-QOL.

SES, measured as the highest completed educational level of the parents either separately or combined. There were some differences in the average SES between the CI and NH group, which might partly be due to differences in recruitment procedure. In contrast to some previous studies (e.g., Kirman & Sari, 2013), we found no association with HR-QOL in

the CI group. In the Kirman and Sari study 26% of the mothers of the children had not completed primary school, and 66% of the families in the study reported that their families had poor economic status (Kirman & Sari, 2013). In contrast, all of the mothers in the current study had completed primary school, and 63% of the mothers in the CI group had completed at least one year of university or college. The SES of the families in the current study is thus very different from some of the articles that report an association between SES and HR-QOL. Furthermore, the educational level in Norway in general is relatively high, and differences in living standard between social classes are fairly small, seen in a global context. Cochlear implantation and habilitation are provided to all individuals that can benefit from this procedure with all costs covered by the Norwegian health care system. The families of children with CIs thus represent the average of the Norwegian population.

Proxy-reported functional hearing in everyday situations (SSQ-index) were weakly to moderately correlated with several of the HR-QOL domains in the present study. This indicated that children who are perceived to hear better in everyday situations seem to have better HR-QOL. In addition, there was a tendency that children who heard better in noise were reported to have higher overall HR-QOL. These findings are in line with previous studies showing that hearing in noise appears to be related to mental health and peer interactions (Huber, 2005; Huber et al., 2015). In the studies by Huber et al. (2005; 2015) the authors argue that children who have difficulties with communication in noise may be at risk of social isolation in school, where much of communication happens in noisy environments. The children who are struggling under these conditions may experience more social isolation, which in turn can lead to problems with mental health over time (Huber et al., 2015).

These findings suggest a need for better listening conditions for the children in our study. Much of the school days are spent communicating in adverse listening conditions, especially for the children who are integrated in mainstream schools, which is the vast majority

of the children in both our study and amongst children with CIs in Norway. In a report by Hendar (2012) on teaching outcomes in children with hearing loss in Norway, only 50% of the teachers report that the children are taught in classrooms that are acoustically adapted and are using technical equipment to enhance listening conditions such as teacher or pupil microphones. Given the results of the current study, with a tendency for children who have better hearing in noise and everyday situations have higher levels of HR-QOL, this might suggest that a better sound environment is important to increase the social inclusion and school functioning in children with CIs. The report by Hendar (2012) suggests that there is substantial room for improvement in this domain in Norwegian schools.

We know little about what *type* of habilitation or support the children in the current study with CIs received during the first years following implantation, or what support they currently receive in school. We do, however, have information about the *amount* of guidance to parents and special education services provided to children as reported by their parents. Though most children in our study did receive additional support in school, the results of our study might suggest that the support the children receive either is not substantial enough, or it is not the right form of support.

The results also indicate that it may not be sufficient to improve the physical listening conditions. There were moderate associations between language abilities and both the HR-QOL Total score and the Psychosocial health composite scale. Additionally, language components such as verbal working memory and expressive language were related to performance on the subscales Social functioning and School functioning. These findings are consistent with the results of previous studies that have identified an association between language abilities and psychosocial functioning or general HR-QOL (Fellinger, Holzinger, Beitel, et al., 2009; Fellinger, Holzinger, Sattel, Laucht, & Goldberg, 2009; G. Leigh et al., 2015; Percy-Smith et al., 2008; Theunissen et al., 2015). From these results, we can derive the need for more specific

follow-up targeting the children's over-all spoken language abilities. As the children are predominantly educated in mainstream schools, mastering the common communication form is the key to peer-interaction. The educational support many of the children receive seem to be insufficient, and there is a need for a more systematical approach to spoken language acquisition from an earlier age and a close monitoring of language skills. In sum, the children who use CIs seem to struggle more with social relations and school functioning than NH children, suggesting that the habilitation and school situation need to be further adjusted to increase these children's HR-QOL.

Strengths and Limitations

As with all studies using generic questionnaires to study HR-QOL, there is a question of content validity. HR-QOL is a particularly ill captured concept, and it can be discussed how well the domains included in the present questionnaire represent the concept. We have, however, used a questionnaire that has been previously been recommended for use in the clinical group we study, and that has been developed with regards to the definition of HR-QOL provided by the WHO. The study could also have been stronger, had responses from the children themselves been included in addition to proxy-report. HR-QOL is generally regarded to be highly subjective, and parents and children may have different perspectives on the same situation as they may emphasize different factors.

We have also examined a specific group of children with CIs, and the results may not apply to all children with CIs nor to children with hearing loss in general. It is, however, also a strength that our sample was composed of a relatively homogenous group of children with bilateral hearing loss and no additional disabilities known to affect neither language development nor HR-QOL. When this group of children was compared to a control group of normal hearing children, the two groups differed mainly on hearing loss and not with regards to a number of confounding variables. Moreover, a number of different language and hearing-

related background variables were systematically examined to assess how these were related to HR-QOL. This is important in order to use the information to review current practice and influence the focus of further habilitation.

Conclusion

Although the overall parent-reported HR-QOL was significantly lower in children with CIs than in children with NH, 57% of the children in the CI group had overall scores within normal limits. The children in the CI group in the current study appear to struggle the most with social interactions and school functioning, and as much as half of the children in the CI group seem to be at risk for impaired HR-QOL in social relations and in school. There were however, no difference between groups on the domains Physical health and Emotional functioning. This is an important finding, as it indicates that the nearest equivalent to mental health in this study is not diminished in the children with CIs. The children who had better language abilities were reported to perform better in both social interactions and in school. Better perceived hearing in different everyday situations was related to better proxy-reported HR-QOL in children with CIs, indicating that the possibility to communicate and hear in everyday environments is important to the overall well-being of the children. This finding highlights the need for better interventions enabling the children to fully participate with their peers in school. Improved habilitation and school interventions have the potential to make a difference for the wellbeing of children with CIs. Such habilitations should focus both on spoken language skills, as well as improving the sound environment, not only in class, but also during breaks. Such interventions will possibly facilitate social interactions with peers and the overall functioning in school for this group of children.

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Figure 1. Boxplots of the distribution and comparison of proxy-reported Pediatric Quality of Life Inventory (PedsQL) in children with cochlear implants (CI) and children with normal hearing (NH). Error bars showing confidence intervals 95%.