Health-related quality of life in South-African children who use cochlear implants

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Abbreviations: CI, cochlear implant; CCIPP, Children with Cochlear Implants: Parental Perspectives; HRQoL, health-related quality of life

ABSTRACT

Objective: To describe health-related quality of life (HRQoL) outcomes of school-going pediatric cochlear implant (CI) recipients in a South-African cohort from the perspectives of parents.

Design: Parents of school-going CI recipients completed the *Children with Cochlear Implants: Parental Perspectives* (CCIPP) CI-specific HRQoL questionnaire. The effect of different demographic variables on HRQoL outcomes was also determined.

Study sample: The study sample included 54 parents of school-going (mean age = 12.2 years; SD = 3.6; range = 6.6 -18.3 years) CI recipients with at least six months CI experience.

Results: Children's communication and general functioning with a CI received the most positive parental ratings. Among a number of confirmed statistically significant ($p < 0.05$) associations between HRQoL outcomes and demographic variables, pre-lingual onset of deafness was linked to better HRQoL in terms of general functioning and well-being. While shorter duration of deafness and unilateral implantation were associated with higher parental ratings for self-reliance and well-being respectively, longer duration of CI use was linked to improved HRQoL outcomes in terms of general functioning.

Conclusion: Parents assigned positive ratings to their child's HRQoL. This exploration of children's HRQoL related to their CIs contributes to evidence-based pediatric CI services that promote optimal psychosocial outcomes.

Keywords: cochlear implant, health-related quality of life, outcomes, parents, pediatric cochlear implantation, quality of life

INTRODUCTION

Cochlear implantation has become a well-established treatment option for children with severe to profound hearing loss who obtain insufficient benefit from acoustic amplification. Not only does cochlear implantation have a positive effect on the language acquisition skills, speech perception, speech production, and communication performance in young children, but it also has a broader impact on their psychosocial well-being (Sack & Whynes, 2005; Warner-Czyz et al, 2013). In recognizing the need to actualize and measure the benefits and limitations of medical interventions such as cochlear implantation on an individual's social, emotional and physical well-being, the term health-related quality of life (HRQoL) has been defined (Loeffler et al, 2010). In order to provide an all-inclusive account of cochlear implant (CI) outcomes for an individual, the functional impact of permanent hearing loss and consequent treatment on that individual's personal well-being should be assessed through HRQoL measures (Zaidman-Zait, 2010). Clinical assessments of CI efficacy typically relate to communication and auditory skills and these measures do not necessarily represent the CI's effect on everyday functioning (Clark et al, 2012; Meserole et al, 2014). As a result, HRQoL has recently become a widespread outcome measure to quantify and monitor the effects of cochlear implantation not only for adults, but also for children.

The impact of a CI on a child's well-being can be measured by using generic or condition-specific HRQoL measures. Condition-specific measures are sensitive to both positive and negative consequences of a specific disorder or treatment, while generic measures focus on broader HRQoL domains such as social well-being and self-esteem, independent of a disorder or medical condition (Hinderink et al, 2000; Bjornson & McLaughlin, 2001; Wiebe et al, 2003; Kumar et al, 2015). Obtaining HRQoL scores for the pediatric CI population

usually involve either the parents (parent-report) or the CI recipients themselves (self-report). Some pediatric CI recipients may not necessarily have the required skills to express their feelings or describe experiences as a result of immature communication skills (Hays et al, 2006). Responses from parents can be used in such cases to describe HRQoL, since parents are considered as the closest proxy to a child. Not only do parents provide valuable information about their child`s general functioning, additional intervention needs and experienced benefits from cochlear implantation (Damen et al, 2007), but they also provide useful insight into their child's socio-emotional and physical well-being (Warner-Czyz et al, 2009).

Significant improvement in HRQoL has been documented for pediatric CI recipients after CI surgery (Beadle et al, 2000; Barton et al, 2006; Sach & Barton, 2007). Positive ratings for a variety of generic and CI specific HRQoL domains have been reported in the literature across HRQoL measures, including communication (Incesulu et al, 2003; Sack & Whynes, 2005; Huttunen et al, 2009; Warner-Czyz et al, 2009; Loy et al, 2010; Fortunato-Tavares et al, 2012; Kumar et al, 2015), general functioning (Archbold et al, 2002; Schorr et al, 2009; Kumar et al, 2015); physical and psychological well-being (Warner-Czyz et al, 2009; Loy et al, 2010; Zhao et al, 2019), social relations (Incesulu et al, 2003; Sack & Whynes, 2005; Huttunen et al, 2009; Fortunato-Tavares et al, 2012; Kumar et al, 2015; Zhao et al, 2019) and self-reliance (Incesulu et al, 2003; Sack & Whynes, 2005; Huttunen et al, 2009; Fortunato-Tavares et al, 2012). However, literature reports less positive ratings for HRQoL domains that relate to education (Soleimanifar et al, 2015; Kumar et al, 2015; Zhao et al, 2018), the effects of the CI on the recipient (Sack & Whynes, 2005; Huttunen et al, 2009; Fortunato-Tavares et al, 2012; Kumar et al, 2015) and the support required by the implanted child (Sack & Whynes, 2005; Huttunen et al, 2009; Fortunato-Tavares et al, 2012).

Various demographic factors have been identified to influence HRQoL ratings in pediatric CI recipients. Female gender, higher levels of parental education, onset of hearing loss at an older age and the absence of additional special needs are associated with more favorable generic HRQoL outcomes in pediatric CI recipients (Barton et al., 2006; Sach & Barton, 2007). Younger chronological age, earlier implantation, longer duration of CI use, higher level of maternal education and lower levels of family stress are also associated with more positive ratings of generic and CI specific HRQoL (Sach & Barton, 2007; Schorr et al, 2009; Warner-Czyz et al, 2009; Warner-Czyz et al, 2011; Meserole et al, 2014; Zhao et al, 2019).

Globally the number of pediatric CI surgeries is steadily increasing due to the expansion of selection criteria, including those children with less severe degrees of hearing loss as well as those with additional medical conditions (Fitzpatrick et al, 2009; Black et al, 2011).Therefore the measurement of HRQoL outcomes has become a fundamental component in pediatric cochlear implantation since it contributes to evidence-based services that eventually endorse optimal outcomes. In spite of this recent focus to assess the broader personal impact of permanent hearing loss and cochlear implantation in children, further investigation is needed to explore the possible influence of different interacting factors on HRQoL outcomes.

At the end of 2017, just more than 1000 children have been implanted at 11 respective CI programs in South Africa (South African Cochlear Implant Group, 2017). A previous report on hearing loss diagnosis and age of intervention of 264 pediatric CI recipients in South Africa, indicated that hearing loss was typically diagnosed late (15.3 months), resulting in delayed initial hearing aid fitting (18.8 months), enrollment in early intervention services (19.5 months) and eventual cochlear implantation (43.6 months) (le Roux et al, 2015). Within

a resource-limited country where developed world benchmarks for pediatric cochlear implantation are difficult to attain, outcome data is critical to guide and influence policy for funding and service provision. Only recently has preliminary data been published on pediatric CI outcomes and predictors of these outcomes within a South African sample of 301 pediatric CI recipients (le Roux et al, 2016). However, HRQoL was not included as an outcome measure and as a result, no published data yet exists on HRQoL outcomes in pediatric CI recipients in South Africa. A number of studies on HRQoL outcomes in pediatric CI recipients reported that parents are less satisfied with their child's education compared to other HRQoL sub-domains (Sack & Whynes, 2005; Huttunen et al, 2009; Kumar et al, 2015; Zhao et al, 2019). In addition, limited available information on educational setting and the inclusion of very young children (not yet in a formal educational setting) have been acknowledged as limitations in recent studies that used a validated CI-specific measures to report on HRQoL in children with CIs (Kumar et al, 2015; Zhao et al, 2019). In an attempt to investigate why parent expectations are often not met in terms of education, this study excluded parents of very young (<6 years of age) CI recipients and gave prominence to parents of older CI recipients who attend formal schooling. Therefore, this study aimed to describe HRQoL outcomes of school-going CI recipients in a South African cohort from the perspectives of parents.

METHODS

Study population

This study included 54 parents of school-going (6-18 years) pediatric CI recipients. Three CI programs in South Africa participated in this multicentre study, from which two programs are situated in the Gauteng Province (Pretoria Cochlear Implant Unit and the Johannesburg

Cochlear Implant Centre) and one program in the KwaZulu-Natal Province (Durban Cochlear Implant Program). Eligible participants for this study included parents of school-going (aged 6-18 years) pediatric CI recipients with at least one CI and with a minimum of six months implant use at the time of data-collection. Only parents who were proficient in English were considered for participation since the validated HRQoL measure used for data collection was used in its original English format. Parents with implanted children who were diagnosed with visual, cognitive or developmental delays were excluded. Demographic and clinical characteristics of 54 pediatric CI recipients are presented in Table 1 and Table 2 respectively.

Materials for data-collection

Parents completed a parental proxy CI specific HRQoL questionnaire, the *Children with Cochlear Implants: Parental Perspectives* (CCIPP) (Archbold et al, 2008). The CCIP is an established HRQoL assessment for children with CIs and validity and reliability of the questionnaire have been confirmed (Nunes et al, 2005; O'Neill et al, 2004). The development of the CCIPP has been fully described (Archbold et al, 2002; O'Neill et al, 2004). Originally the CCIPP was employed as a parent-directed retrospective interview (Archbold et al, 2002). Demonstrating consistent results across methodologies, subsequent studies made use of semi-structured interviews (Nunes et al, 2005), and also self-administered surveys (Incesulu et al, 2003; O'Neill et al, 2004; Archbold et al, 2008; Huttunen et al, 2009; Kumar et al, 2015; Zhao et al, 2019). It includes 74 statements in which two main domains of the cochlear implantation process is covered: decision-making (26 items) and outcomes of implantation (48 items). Eight sub-domains are used to categorize the outcomes of implantation, consisting of six child-related sub-domains (*communication, general functioning, well-being, self-reliance, social relations* and *education*) and two family-related sub-domains (*effects of*

implantation and *supporting the child*). Items were randomized in order to avoid a halo effect in responses to items referring to the same domain. Parents were expected to rate their responses to each statement on a 5-point Likert scale (ranging from strongly disagree to strongly agree). Most (46) statements are worded positively and 28 are worded negatively in order to balance items for negativity and positivity. Only recently, CI specific child and parent-proxy HRQoL measures were developed for children (ages 6 to 12) using the Food and Drug Administration Guidance (2009) on patient-reported outcomes (Hoffman et al, 2019). However, the CCIPP was the only CI specific parent-report HRQoL measure that was available at the time of data collection for this study.

Additional demographic, hearing loss and CI related data were captured for the purpose of this study. In conjunction to completing the CCIPP questionnaire, parents provided basic demographic information, while additional retrospective CI and hearing loss related data were captured from clinical patient files by CI team coordinators.

Data collection

Institutional ethics committee approval was obtained before data collection commenced. Parents who met the inclusion criteria of this study were contacted by telephone or were approached during clinical consultations by the researchers (authors 1-4) to inform them about the purpose and procedures of the study. Those parents willing to participate received an information letter containing the purpose of the study and requirements for participation. Informed consent was required from each parent participant prior to the initiation of data collection. A total of 120 questionnaires were distributed among the three participating CI programs and 54 questionnaires were returned (response rate of 45%). Parents who consented

to participate were requested to complete the CCIPP questionnaire either electronically (online) or in hard-copy, within seven working days of receiving the questionnaire. Data were collected over a four months data collection period.

Statistical analysis

A commercially available statistical software package (SAS version 9.4) was used for the analysis. The eight domains outlined in the overarching domain of outcomes of implantation in the CCIPP were used for the HRQoL analysis in this study. Values were assigned to responses obtained on a 5-point Likert scale (1 = strongly disagree to 5 = strongly agree), making it possible to calculate standard deviations and estimate the dispersion in scores (O'Neill et al., 2003). Non-responses were processed as missing values. Scoring of negative statements were reversed in order for higher values to correspond to a more positive response that represent better HRQoL (Huttunen et al, 2009; Kumar et al, 2015; Zhao et al, 2019). Scores for each HRQoL domain were averaged to yield a domain and overall HRQoL mean for each participant.

Frequency tables and descriptive statistics were used to describe the implanted children of parent participants in terms of demographic, CI and hearing loss characteristics (Tables 1 and 2). The criteria used to differentiate between pre- and post-lingual deafness in pediatric CI recipients was the age at diagnosis of severe-profound hearing loss before and after three years of age respectively (Dowell et al, 2004; Ruffin et al, 2013). All 26 children with bilateral implants whose parents participated had at least six months experience with their bilateral implants at the time of data collection.

Distribution-free nonparametric statistics were used to accommodate for the smaller sample size and the use of ordinal data. Multiple comparisons of HRQoL domains were done by means of post hoc testing. By using the Wilcoxon signed-rank test, pairwise comparisons were performed to determine if significant differences exist between HRQoL domains. In order to determine if associations exist among the respective HRQoL sub-domains, Spearman correlation coefficients were calculated.

Spearman correlation coefficients were also used to assess possible associations between continuous variables and HRQoL sub-domain and overall ratings. For the *age at diagnosis of hearing loss/ deafness* and the *age at first CI activation* variables, only the children with congenital/ early-onset hearing loss were considered (n = 40), in order to reflect the current status of early hearing detection and intervention services in South Africa. In order to investigate the influence of categorical variables on HRQoL sub-domain and overall ratings, general linear models were constructed.

Statistical significance was accepted at the traditional $p < 0.05$ level. A Bonferonni adjustment was performed to reduce the likelihood of Type 1 error owing to multiple comparisons.

RESULTS

Comparisons among health-related quality of life domains

All eight HRQoL domains' mean ratings together with the mean rating of the overall HRQoL score (M = 3.79; SD = 0.37; range = 2.60-4.46) surpassed three on a 5-point Likert scale,

demonstrating that parents regarded their child's HRQoL as being more positive than negative.

Table 3 presents descriptive statistics for sub-domain and overall HRQoL ratings of the CCIPP questionnaire. Highest mean scores were obtained for *communication* (M = 4.15, SD = 0.62, range = 2.60-4.46) and *general functioning* (M = 4.05, SD = 0.51, range = 2.83-5.00). Lowest mean scores were obtained for *effects of implantation* (M = 3.49, SD = 0.62, range = 2.00-4.83) and *supporting the child* (M= 3.46, SD = 0.74, range = 2.17- 4.33).

Friedman's analysis of variance indicated a significant main effect of domain on HRQoL ratings (X^2 = (86.353), p < 0.001). Post-hoc testing was employed to compare differences among domains. Both *communication* and *supporting the child* had the highest number of significant interdomain differences.

C*ommunication* obtained significantly more positive ratings than *well-being* ($X^2(1)$ = 4.046, p = 0.001*), social relations* ($X^2(1)$ = 3.673, p = 0.007*) and education* ($X^2(1)$ = 4.950, p < 0.000*). General-functioning* obtained significantly more positive ratings than *education* ($X^2(1)$ = 3.477, p= 0.014*). Effects of implantation* received significantly less positive ratings than *communication* (X^2(1) = 6.737, p < 0.000), *general functioning* (X^2(1) = 5.264, p < 0.000) and *self-reliance* (X^2(1) = 3.752, p < 0.005). Parents rated *supporting the child* significantly less positively than *communication* (X^2(1) = 7.248, p < 0.000), *general functioning* (X^2(1) = 5.775, p < 0.000), *self-reliance* (X^2(1) = 4.262, p = 0.001), *social relations* (X^2(1) = 3.575, p = 0.010) *and well-being* (X^2(1) = 3.202, p < 0.038).

A comparison of descriptive data among two other similar studies is shown in Supplementary Appendix A (Table A1), comparing HRQoL outcomes obtained in this current study to those obtained in the recent studies of Kumar et al (2015) and Zhao et al (2019).

Associations among health-related quality of life sub-domains

In order to determine if associations exist among the respective HRQoL sub-domains, Spearman correlation coefficients were calculated (Supplementary Appendix A, Table A2). Effect size of associations were classified as either small^{+} ($0.10 \geq r < 0.30$), medium^{++} ($0.3 \geq r < 0.50$) or large^{+++} ($r \geq 0.50$) (Cohen, 1988). Correlations are listed in order from greatest to least magnitude. *Communication* achieved the highest number of significant ($p < 0.05$) interdomain associations. Ratings of *communication* positively correlated with five domains, namely *social relations* ($r = 0.65^{+++}$), *self-reliance* ($r = 0.59^{+++}$), *general-functioning* ($r = 0.53^{+++)}$, *education* ($r = 0.53^{+++}$) and *well-being* ($r = 0.52^{+++}$). Positive correlations that were significant ($p < 0.05$) were found between *well-being* and *education* ($r = 0.52^{+++}$), *self-reliance* and *education* ($r = 0.53^{+++}$) and *self-reliance* and *social relations* ($r = 0.37^{++}$).

General functioning achieved significant ($p < 0.05$) positive correlations with three domains, namely *self-reliance* ($r = 0.42^{++}$), *social relations* ($r = 0.35^{++}$) and *well-being* ($r = 0.28^{+}$). Likewise, *well-being* positively correlated significantly ($p < 0.05$) with *self-reliance* ($r = 0.42^{++}$) and *social relations* ($r = 0.35^{++}$). Significant ($p < 0.05$) positive correlations were also found between *social relations* and *education* ($r = 0.42^{++}$), *education* and e*ffects of implantation* ($r = 0.33^{++}$), *social relations* and *effects of implantation* ($r = 0.29^{+}$), as well as *effects of implantation* and *supporting the child* ($r = 0.27^{+}$).

Associations between continuous demographic variables and health-related quality of life ratings

In order to assess associations between continuous demographic variables and HRQoL sub-domain and overall ratings, Spearman correlations coefficients were computed. Five continuous variables were included in the analyses namely age at study (chronological age), age at first CI activation, duration of CI use, age at diagnosis of deafness/severe to profound hearing loss and duration of deafness prior to cochlear implantation (Table 4). Yet again, effect size of associations were classified as either small^{+} ($0.10 \geq r < 0.30$), medium^{++} ($0.3 \geq r < 0.50$) or large^{+++} ($r \geq 0.50$) (Cohen, 1988). A significant positive correlation was found between *duration of CI use* and the *general functioning* domain, implying that parents of children who had longer CI experience assigned more positive ratings for their children's general functioning ($r = 0.31^{++}$; $p = 0.024$). In contrast, a significant negative correlation was found between *duration of CI use* and *effects of implantation*, indicating that parents of children who had longer CI experience were less satisfied with the *effects of implantation* relative to parents of children who had less CI experience ($r = -0.33^{++}$, $p = 0.014$). *Duration of deafness* correlated negatively with the *self-reliance domain*, suggesting that parents of children who had a longer duration of deafness prior to cochlear implantation, assigned lower ratings to their children's self-reliance relatively to parents of children who had a shorter duration of deafness prior to implantation ($r = -0.31^{++}$, $p = 0.022$).

Associations between categorical demographic variables and health-related quality of life ratings

A general linear model was constructed to investigate the influence of categorical demographic variables on overall HRQoL ratings. Likewise, general linear models were also

constructed for each of the HRQoL sub-domains. Five categorical variables were considered for the modeling and included *gender* (male/female), *educational setting* (mainstream/ non-mainstream), *cochlear implantation* (unilateral/ bilateral), *onset of deafness* (pre-/ post-lingual) and *highest level of parental education* (tertiary/ high school or matric). The simultaneous effect of these categorical independent variables was measured for HRQoL outcomes and therefore the influence of an independent variable is significant on the outcome in the presence of the other independent variables.

No significant associations were found between categorical demographic variables and overall HRQoL ratings. Table 5 presents the general linear regression analysis results in terms of the four HRQoL sub-domains that yielded significant ($p<0.05$) associations with categorical demographic variables. Significant associations were only obtained for the *communication, general functioning, well-being* and *education* sub-domains (Table 5).

Educational setting was significantly associated with both the *communication* ($p = 0.037$) and *education* ($p = 0.016$) sub-domains. Parents of children in mainstream educational settings on average scored their childrens' communication abilities (M = 4.33, SD = 0.54) and general performance in school (M = 3.93, SD = 0.58) higher than parents whose children are not in mainstream educational settings (M = 3.98, SD = 0.66; M = 3.48. SD = 0.62).

Onset of deafness was significantly associated with the *general functioning* ($p = 0.021$) and *well-being* ($p = 0.047$) sub-domains, indicating that parents whose children were pre-lingually deafened on average score their children's general functioning (M = 4.16, SD = 0.45) and well-being (M = 3.88, SD = 0.60) higher than those parents whose children were post-lingually deafened (mean = 3.71, SD = 0.57; mean = 3.61, SD = 0.57). Cochlear implantation was also significantly associated with the *well-being sub-domain* ($p = 0.035$),

signifying that parents of children implanted bilaterally on average score their children's well-being less postively (M = 3.66, SD = 0.61) in comparison to parents of children implanted unilaterally (M = 3.94, SD = 0.57).

DISCUSSION

Parents of school-going pediatric CI recipients in this study perceived their children's HRQoL to be more positive than negative, by assigning positive ratings (on average exceeding three on a sub-scale of five) to overall HRQoL and all HRQoL sub-domains. Post-operative average ratings exceeding three out of five for all eight HRQoL sub-domains of the CCIPP have been confirmed by a number of studies (Huttunen et al, 2009; Kumar et al, 2015; Zhao et al., 2019).

The *communication* sub-domain (referring to the ease, quality, and quantity ofcommunication and conversation of the implanted child) achieved the highest mean score and parents rated this sub-domain significantly more positively than all other sub-domains, except for *general functioning* and *self-reliance*. It is clear from this study and from reports formerly published that parents consider their child's communication as one of the most advantageous aspects of HRQoL post-implantation (Incesulu et al, 2003; Sack & Whynes, 2005; Huttunen et al, 2009; Warner-Czyz et al, 2009; Loy et al, 2010; Fortunato-Tavares et al, 2012; Kumar et al, 2015). The highest number of significant inter-domain associations was also achieved for the *communication* sub-domain, indicating positive correlations between children's communication and their *social relations*, *self-reliance*, *general-functioning*, *education* and *well-being*. According to parent perceptions, this implies that improved communication abilities in children with CIs also lead to better relationships, greater independence, more

reliance on auditory information, steady progress in academics and improved overall happiness.

The second highest mean score was obtained for the *general-functioning* sub-domain (reflecting changes in attention, safety and engagement of the implanted child). Overall, parents perceived their child's increased dependence on auditory information and functional hearing in everyday environments as a positive outcome of implantation (Huttunen et al, 2009). Significant positive correlations were indicated between the *education* (referring to the performance of the child at school, as well as placement and responsiveness within the educational setting) and *well-being* (indicating the happiness and frustration of the implanted child) sub-domains, as well as the *education* and *self-reliance* (signifying the child's confidence and independence) sub-domains. In agreement with previously published reports (Sack & Whynes, 2005; Huttunen et al, 2009; Kumar et al, 2015), the *education* sub-domain on average received modest parental ratings in this study. Irrespective, higher parental satisfaction with education strongly influenced overall well-being and independence.

The two family-related sub-domains, namely *effects of implantation* (reflecting the child's progress with the CI, future concerns regarding device functioning, and the child's reaction to the device) and *supporting the child* (referring to the amount and effects of help required by the child before and after implantation) received least positive ratings by parents in this study. Parents rated *supporting the child* significantly less positively than nearly all other sub-domains, except for *education* and *effects of implantation*. Lower ratings for *effects of implantation* (Huttunen et al, 2009; Schorr et al, 2009; Almeida et al, 2015; Kumar et al, 2015) as well as *supporting the child* (Huttunen et al, 2009; Almeida et al, 2015) were also

obtained in other studies on parent-perceived HRQoL in pediatric CI recipients. These findings indicate that parents, in general, are not only concerned about their child's future device function and reactions to the CI but also about the assistance their child will require in everyday life of the family. Children receiving CIs are affected in not only their own HRQoL, but also in that of the whole family (Beadle et al, 2000). Parents need to cope with the changes that occur in the family's dynamics during and after implantation, and this process requires adaptation (Allegretti, 2002). CI professionals should ensure that families have access to a range of formal and informal individualized support systems that will facilitate family adjustments and enhance the health and well-being of the family unit (Moeller et al, 2013). The more negative ratings of family-related HRQoL in this study could possibly point to poor connections to required support systems that enable families to accrue the necessary knowledge and experiences that can enable them to function effectively on behalf of their implanted child (Moeller et al, 2013).

In addition to describing CI-specific HRQoL outcomes from the perspectives of parents, this study also related HRQoL outcomes to a broad range of demographic, hearing loss and CI related variables. Duration of CI use correlated positively with the *general functioning* HRQoL sub-domain, indicating that parents of children who had longer CI experience assigned higher ratings to their child's general functioning in terms of positive changes in attention, safety and engagement (functional hearing). Using an ad hoc, self-reported CI-specific questionnaire, Schorr et al (2009) also indicated that a longer duration of CI use correlated with higher overall HRQoL in a group of 37 pediatric CI recipients (ages 5-14 years). Zhao et al (2019) also confirmed that duration of CI use correlated positively with not only the *general functioning* HRQoL sub-domain of the CCIPP, but also the *communication, self-reliance, well-being* and *effects of implantation* sub-domains. In contrast, a significant

negative correlation was found between duration of CI use and the *effects of implantation* sub-domain in this study. This negative correlation suggests that parents of children who had longer CI experience were less satisfied with the effects of implantation relative to parents of children who had less CI experience. More positive HRQoL with less CI experience was also confirmed by Warner-Czyz et al (2009) for a group of pre-school (ages 4-7 years) CI recipients. With a broader range of ages (6.8 - 18.3 years) and duration of CI experience (0.7 – 15.8 years) in this study, this finding could suggest that the positive effects of implantation become less apparent to parents the longer the child is implanted. Since parents over time become more concerned about their child's future device function, involved professionals should offer on-going counselling and sustained support to parents for extended periods of time after implantation.

The negative correlation found between duration of deafness and the *self-reliance* HRQoL sub-domain indicates that parents of children who had a shorter duration of deafness prior to cochlear implantation assigned better ratings to their child's *self-reliance* (independence and confidence) relatively to parents of children with longer duration of deafness prior to implantation. Since the duration of deafness/ severe-to-profound hearing loss implies the duration of auditory deprivation prior to cochlear implantation, it is known to be a critical predictor of implantation success (le Roux et al, 2017). Therefore, study results suggest that the shorter the period of auditory deprivation prior to implantation is, the more confident and independent parents will perceive their children to be.

The *communication* sub-domain, as well as the *education* sub-domain were significantly associated with educational setting. These results imply that parents of children in

mainstream educational settings on average scored their children's communication abilities and school performance higher than parents of children in non-mainstream educational settings. With the majority (79.63%) of the parents' children in this study sample using spoken language for communication and with almost half (48.15%) of the children being in mainstream educational settings, the association between educational setting and parental ratings for the *communication* sub-domain should be interpreted with caution. It is most likely that pediatric CI recipients with better spoken language and communication abilities would rather be integrated into mainstream educational settings than non-mainstream settings. Even though mainstream education is likely to be a realistic outcome for children implanted early and without additional developmental difficulties (Archbold et al, 2002; Damen et al, 2007), the emphasis of educational placement for pediatric CI recipients should rather be on the appropriateness of the educational setting to each child's specific needs (le Roux et al, 2016). Also, it is to be expected that parents of CI recipients with poorer spoken language and communication abilities post-implantation enroll their children in non-mainstream educational settings, and rate HRQoL less positively because they might have more concerns for their child's future based on poorer communication abilities. Irrespective, the relationship between mainstream schooling and parent satisfaction with educational placement, performance, and progress was evident in this study.

Within this study sample, the onset of deafness had a significant influence on both *general functioning* and *well-being* HRQoL sub-domains. Parents of prelingually deafened children on average perceived their children's general functioning with the help of hearing and their overall well-being and happiness to be more positive than parents of post-lingually deafened children. This could relate to the fact that earlier age at onset of deafness concur with better psychosocial adjustment in school-going deaf children (Polat, 2003).

Contrary to expectations, parents of children with bilateral CIs on average scored their children's *well-being* more negatively in comparison to parents of children implanted unilaterally. However, this association between bilateral implantation and HRQoL outcomes was only evident for one of the HRQoL sub-domains, and not for overall HRQoL outcomes. Irrespective, this finding is in contrast to a number of studies confirming the strong association between bilateral implantation and improved HRQoL outcomes in adult (Olze et al, 2012; Härkönen et al, 2015; le Roux et al, 2017) and pediatric (Samuel et al, 2016) CI recipients. Evidence on whether bilateral implantation significantly improve broader outcomes such as HRQoL, is lacking for pediatric CI recipients (Johnston et al, 2009; Sparreboom et al, 2010). This study therefore provides some evidence that the expectations of parents of bilaterally implanted children are not necessarily met in terms of the overall comfort and happiness of their children. Also, since family financial resources remains a decisive factor for bilateral implantation in South Africa, not all children have equal opportunity to access a second implant (le Roux et al, 2016). It could be that the parental expectations of this selective (more privileged) sub-group of bilateral pediatric CI recipients in this study are very high and not easily met. It should however be noted that only three of the eleven CI programs in South Africa participated in this multicenter study, resulting in a relatively small sample size. Consequently the study sample for this research could not be considered as representative of parents of pediatric CI recipients in South Africa and results should not be generalized.

CONCLUSION

Parents of school-going pediatric CI recipients in this study assigned positive ratings to overall HRQoL and all HRQoL sub-domains. A mainstream educational setting was

associated with better HRQoL outcomes in terms of communication and education, while pre-lingual onset of deafness was associated with better HRQoL outcomes in terms of general functioning and well-being. Longer duration of deafness and bilateral implantation were associated with lower parental ratings for self-reliance and well-being respectively. Longer duration of CI use was linked to better ratings for general functioning, but poorer ratings for effects of implantation. This study provided valuable insights into parental perceptions of CI outcomes in terms of HRQoL. This investigation of children's HRQoL associated with their CIs contributes to evidence-based pediatric CI services that promote optimal psychosocial outcomes and assist professionals to make the best decisions about the required care and support for pediatric CI recipients and their parents.

Acknowledgments

The authors acknowledge the support of involved professionals from the three participating CI programs (Pretoria Cochlear Implant Unit, Johannesburg Cochlear Implant Centre and Durban Cochlear Implant Program) who contributed towards data collection for this study. Andries Masenge is to be acknowledged for his guidance and valuable support with the data analysis.

Declaration of interest

The authors declare no conflicts of interest.

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Table 1. Demographic characteristics of school-going pediatric cochlear implant recipients *(n =54)*

Demographics	***% (n)***
Age at study *(years)*	
Mean (SD)	12.21 (3.60)
Range	6.60 - 18.30
Gender *	
Male	53.70 (29)
Female	46.30 (25)
Educational setting *	
Mainstream private school	27.78 (15)
Mainstream public school	20.37 (11)
Special needs school	18.52 (10)
School for the Deaf (South-African Sign Language mode of communication)	16.67 (9)
School for the hard of hearing (oral mode of communication)	9.26 (5)
Homeschool	7.41 (4)
Communication mode *	
Spoken language	79.63 (43)
Total communication (South African Sign Language and spoken language)	20.37 (11)
Highest level of parental education *	
Tertiary qualification (University or other)	62.96 (34)
Matric completed	20.37 (11)
High school (Gr8-11)	14.82 (8)
Primary school (Gr1-7)	1.85 (1)

* *parent-reported data*
** *only congenital/ early onset hearing loss were considered (n = 40)*

Table 2. Hearing loss and cochlear implant characteristics of school-going pediatric CI recipients *(n = 54)*

Hearing loss and cochlear implant characteristics	***% (n)***
Unilateral/ bilateral cochlear implantation *	
2 cochlear implants (bilateral)	48.15 (26)
1 cochlear implant with hearing aid in the non-implanted ear (bimodal)	31.48 (17)
1 cochlear implant without a hearing aid in non-implanted ear	20.37 (11)
Age at first cochlear implant *(years) (n=40)***	
Mean (SD)	3.90 (2.41)
Range	0.62 - 11.60
Age at second cochlear implant (*years) (n = 26)*	
Mean (SD)	6.60 (3.90)
Range	1.50 - 13.70
Time-lapse between first and second implant *(years)* (n =26)	
Mean (SD)	3.34 (3.04)
Range	0.20 - 10.60
Duration of CI use *(years)*	
Mean (SD)	8.21 (4.10)
Range	0.70 - 15.80
Age at diagnosis of hearing loss (*months)* **	
Mean (SD)	19.83 (12.62)
Range	1.00 - 54.00
Age at diagnosis of deafness/severe to profound hearing loss *(months)* **	
Mean (SD)	20.38 (14.34)
Range	1.00 - 60.00
Duration of deafness prior to cochlear implant *(years)*	
Mean (SD)	1.90 (2.00)
Range	0.10 - 9.70
Onset of hearing loss	
Congenital/early onset	74.07 (40)
Unknown	16.67 (9)
Acquired/ progressive/ sudden onset	9.26 (5)

* *parent-reported data*

** *only congenital/ early onset hearing loss were considered (n = 40)*

Table 3. Health-related quality of life scores depicted from *CCIPP* results *(n = 54)*

CCIPP questionnaire	***Description of sub-domain****	***Mean (SD)***	***Median***	***Range***
Total HRQoL score		3.79 (0.37)	3.77	2.60-4.46
CCIPP sub-domains				
Communication	Ease, quality, and quantity of communication and conversation	4.15 (0.62)	4.14	2.00-5.00
General Functioning	Changes in attention, safety, and engagement	4.05 (0.51)	3.92	2.83-5.00
Well Being	Happiness and frustration	3.81 (0.60)	3.80	2.40-5.00
Self-Reliance	Indicators of confidence and independence	3.88 (0.63)	4.00	2.00-5.00
Social Relations	Relationships within and outside the family	3.87 (0.52)	3.86	2.71-5.00
Education	Performance of the child at school; placement and responsiveness within the school district	3.70 (0.64)	3.71	2.29-5.00
Effects of implantation	Progress with the cochlear implant, future concerns regarding device function, and child reaction to the device	3.49 (0.62)	3.54	2.00-4.83
Supporting the child	Amount and effects of help required by child before and after implantation	3.46 (0.47)	3.50	2.17-4.33

**Description according to Archbold et al., 2008; Kumar et al., 2015*

Table 4. Associations between continuous demographic variables and health-related quality of life ratings

Health-related quality-of-life sub-domain	**Age at study** (n = 54)	**Age at first CI activation** (n = 40)	**Duration of CI use** (n = 54)	**Age at diagnosis of deafness** (n = 40)	**Duration of deafness** (n = 54)
Communication	0.08	-0.31	0.14	-0.19	-0.16
General functioning	0.09	-0.25	0.31*	-0.14	-0.23
Well-being	-0.20	-0.03	-0.13	-0.15	0.00
Self-reliance	-0.03	-0.31	0.12	-0.15	-0.31*
Social relations	-0.17	-0.08	-0.16	-0.14	0.01
Education	-0.21	-0.16	-0.22	-0.08	-0.03
Effects of implantation	-0.19	-0.23	-0.33*	0.24	0.13
Supporting the child	0.00	-0.01	0.04	-0.01	-0.12
Overall HRQoL	-0.15	-0.17	-0.09	-0.13	-0.10

Spearman correlation coefficient

** $p < 0.05$*

Table 5: Associations between categorical demographic variables and health-related quality of life sub-domain ratings *(n = 54)*

Health-related quality of life subdomain	**Categorical demographic variable**	**Pr>F (p-value)**	**Categories**	**Mean score (SD)**
Communication	Education setting	0.0374	Mainstream (n = 20)	4.33 (SD: 0.54)
			Non-mainstream (n = 34)	3.98 (SD: 0.66)
General functioning	Onset of deafness	0.0214	Pre-lingually deafened (n = 40)	4.16 (SD: 0.45)
			Post-lingually deafened (n = 14)	3.71 (SD: 0.57)
Well-being	Cochlear implantation	0.0345	Bilateral (n = 26)	3.66 (SD: 0.61)
			Unilateral (n = 28)	3.94 (SD: 0.57)
	Onset of deafness	0.0465	Pre-lingually deafened (n = 40)	3.88 (SD: 0.60)
			Post-lingually deafened (n = 14)	3.61 (SD: 0.57)
Education	Educational setting	0.0161	Mainstream (n = 26)	3.93 (SD: 0.58)
			Non-mainstream (n = 28)	3.48 (SD: 0.62)