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Health-Related Quality of Life in Children With Low Language or Congenital Hearing Loss, as Measured by the PedsQL and Health Utility Index Mark 3

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ABSTRACT

Objectives: To examine health-related quality of life (HRQoL) in young children with low language or congenital hearing loss and to explore the value of assessing HRQoL by concurrently administering 2 HRQoL instruments in populations of children.

Methods: Data were from 2 Australian community-based studies: Language for Learning (children with typical and low language at age 4 years, n = 1012) and the Statewide Comparison of Outcomes study (children with hearing loss, n = 108). HRQoL was measured using the parent-reported Health Utilities Index Mark 3 (HUI3) and the Pediatrics Quality of Life Inventory 4.0 (PedsQL) generic core scale. Agreement between the HRQoL instruments was assessed using intraclass correlation and Bland-Altman plots.

Results: Children with low language and with hearing loss had lower HRQoL than children with normal language; the worst HRQoL was experienced by children with both. The lower HRQoL was mainly due to impaired school functioning (PedsQL) and speech and cognition (HUI3). Children with hearing loss also had impaired physical and social functioning (PedsQL), vision, hearing, dexterity, and ambulation (HUI3). Correlations between instruments were poor to moderate, with low agreement.

Conclusions: Children with low language and congenital hearing loss might benefit from interventions targeting overall health and well-being, not just their impairments. The HUI3 and PedsQL each seemed to provide unique information and thus may supplement each other in assessing HRQoL of young children, including those with low language or congenital hearing loss.

Keywords: children, health-related quality of life, low language and hearing loss.

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Introduction

Health-related quality of life (HRQoL) has become important in the evaluation of resource allocation in healthcare.[1] It is becoming more common as an outcome measure in clinical trials and patient care[2] because of its utility in guiding patient treatment and in population health intervention design. Although childhood language and hearing problems are relatively common, studies of HRQoL in these groups are limited. According to 2 recent reviews, only 7 and 5 studies, respectively, have reported on HRQoL in children with low language (LL) ability[3] and hearing loss.[4] In both cases, most of the reviewed studies included children older than 6 years, and HRQoL was primarily assessed by non–preference-based instruments. Furthermore, because results were mixed, stable conclusions could not be reached on the impact of LL and hearing loss on HRQoL during childhood. Thus, there is a gap in the literature regarding HRQoL in children with LL and hearing loss, especially in children aged 6 years and younger.

A number of generic (preference-based and non–preference-based instruments) and disease-specific instruments have been used to capture HRQoL in adult and pediatric populations.[5] Nevertheless, the choice of HRQoL instruments for trials or research studies is not straightforward because different HRQoL instruments have various purposes at the clinical and population

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level. Generic instruments have the advantage of being applicable to a wide range of populations and conditions, thereby accommodating comparison across populations.[6] Specific instruments, which focus on 1 particular disease or health condition, may be more sensitive to the specific condition and therefore more suitable for use within particular patient groups or populations (for example, when testing new interventions or quality improvement initiatives).[2,6] Both preference-based and non–preference-based instruments reflect individuals' levels of HRQoL based on their perceived health-related functioning and well-being, whereas preference-based instruments also take into account the individuals' preferences for specific health-related states.[7] For economic evaluations of health interventions, preference-based instruments are preferable as they accommodate the calculation of quality-adjusted life-years, which is needed in cost-utility analysis, a technique commonly used to assess resource allocation in healthcare.[2] Nevertheless, the construction of preference-based instruments permits only a brief assessment of the underlying HRQoL aspects, whereas non–preference-based instruments allow for a more extensive assessment of these underlying aspects. Thus, it may be beneficial to use both preference-based and non–preference-based HRQoL instruments when evaluating health interventions. Nevertheless, there have been few attempts to compare the value of assessing HRQoL concurrently by non–preference-based and preference-based instruments.

We aimed to (1) examine the HRQoL in young children with LL, congenital hearing loss (CHL), or both and (2) investigate the complementary value of assessing HRQoL in these 2 populations, and in young children in general, by a concurrent use of 2 established generic instruments, the Pediatric Quality of Life Inventory (PedsQL) and the Health Utilities Index Mark 3 (HUI3). This study will contribute to the dearth of existing literature by reporting HRQoL in young children with LL and/or CHL and providing a comprehensive direct comparison of HRQoL assessed by the PedsQL and the HUI3 in these specific children populations.

Methods

Sample

This article draws on data from the families and children who took part in the Language for Learning trial (L4L)[8,9] and the Statewide Comparison of Outcomes study (SCOUT).[10] Methods and results for both L4L and SCOUT have been reported elsewhere.[8-10] In this article, we use the term LL to refer to children whose language performance falls below well-recognized cut points regardless of known or unknown etiology. Briefly, the L4L study was a 2-armed randomized controlled trial that aimed to improve language and related outcomes in children with LL before school entry. Data for the current article were drawn from the assessment completed at baseline in a community sample (N = 1464), which was representative of 4-year-old children born in Victoria, Australia, in 2006.[8,9] From here on, this whole population is referred to as a general population sample. The SCOUT study was a quasi-randomized trial performed as a natural experiment. This study targeted all 5- to 6-year-old children born with CHL during March 2003 to February 2005 in 2 Australian states: New South Wales and Victoria.

Both L4L and SCOUT were approved by the Royal Children's Hospital Ethics Committee, and all parents provided written informed consent.

Participants and Setting

Participants were recruited to the L4L trial from 7 local government areas in greater Melbourne, Australia. Parents who consented to take part completed a brief questionnaire, and the children received a 1.25- to 1.5-hour formal language assessment around their fourth birthday. Children were classified as having LL if they had expressive and/or receptive language scores more than 1.25 SD below the normative mean on the Clinical Evaluation of Language Fundamentals–Preschool 2 (CELF-P2).[11,12] This subsample of the L4L is hereafter referred to as children with LL. Exclusion criteria included known intellectual disability, autism spectrum disorder, major medical conditions, hearing loss >40 decibels in the better ear, or parents with insufficient English to participate.

Participants in the SCOUT study were recruited from the Australian Hearing database between July 2008 and November 2009.[10] Children were eligible for the study if they had bilateral CHL of >25 decibels pure-tone average (dB HL) in the better ear and were fitted with hearing aids and/or cochlear implants by 4 years of age.[10] This sample is referred to as children with CHL. Parents with eligible children were invited to express their interest and were then provided a 2-hour home visit by a speech pathology or psychology researcher. Children in the SCOUT study were classified as having LL if they had receptive and expressive language scores more than 1.25 SD below the normative mean on the Preschool Language Scale–4 Australian Language Adaption.[13] Children were excluded from the study if their hearing loss was unilateral, acquired (as judged by Australian hearing records), and/or conductive; if their hearing was currently in the normal range; or if their parents had insufficient English to participate.

The analyses in this current study included participants (N = 1120) from the 2 cohorts (L4L baseline cohort, n = 1012, and SCOUT, n = 108) who had both HUI3 and PedsQL data.

HRQoL Measurement

HRQoL was captured by 2 generic instruments, the HUI3 and the PedsQL. The HUI3 captures 8 domains (vision, hearing, speech, ambulation, dexterity, emotion, cognition, and pain/discomfort), with 1 to 2 items per domain and 4 to 6 response alternatives per item.[14] Together, the 12 items generate an overall HRQoL utility score. We used the parent-proxy HUI3 data for the analyses. The HUI3 has demonstrated good discriminant validity and high test–retest reliability (intraclass correlation coefficient of 0.77).[15]

The PedsQL 4.0 generic core scales consist of 4 domains: physical functioning, emotional functioning, social functioning, and school functioning.[16] Combining all items generates an overall HRQoL score. The PedsQL has demonstrated a high level of internal and external reliability (alpha coefficients of total scale scores ≥0.9 for both child self-report and parent-proxy report).[17] In L4L, we used the 21-item PedsQL parent-proxy form for 2- to 4-year-olds, and in SCOUT, we used the 5- to 7-year-old 23-item parent-proxy form, which included 2 additional school functioning items that were not relevant for younger children.

HRQoL Scores

The HUI3 is scored using single- and multidomain utility functions and is reported as a single overall utility score, which is usually presented on a scale between 1 (HRQoL equivalent to full health) and 0 (HRQoL equivalent to being dead), with the lowest possible score being −0.36 (worse than dead).[18] We used preference weights from a Canadian population, as Australian population-specific preference weights have not been generated. The commonly used minimum clinically important difference on the −0.36 to 1 scale is 0.03.[19]

The PedsQL generic core scores are generated on a scale from 0 to 100. A mean score is generated by summing all the items and dividing by the total number of items answered for each scale, with higher scores indicating better HRQoL.[20] A change of 4.5 in

Table 1. Comparison of health-related quality-of-life scores depending on language or hearing ability in children included in the Language for Learning (L4L) community-based sample and the Statewide Comparison of Outcomes (SCOUT) study.*

Characteristic	n (%)	Community-based cohort (L4L, n = 1012)			
		HUI3		PedsQL	
		Mean (SD)	Median (IQR)	Mean (SD)	Median (IQR)
Whole sample	1012 (100)	0.90 (0.13)	0.95 (0.85-1)	85.22 (10.13)	86.90 (79.76-92.86)
Hearing loss severity					
Normal (≤25)	n/a	n/a	n/a	n/a	n/a
Mild	n/a	n/a	n/a	n/a	n/a
Moderate	n/a	n/a	n/a	n/a	n/a
Severe (61-80)	n/a	n/a	n/a	n/a	n/a
Profound (81+)	n/a	n/a	n/a	n/a	n/a
P value					
Language ability					
Typical	886 (87.55)	0.91 (0.13)	0.95 (0.86-1)	85.47 (9.74)	86.90 (79.76-92.86)
LL	126 (12.45)	0.85 (0.15)	0.88 (0.76-1)	83.49 (12.46)	84.52 (76.19-92.86)
P value		<**.001**		0.25	
Mainstream school/ education program	n/a	n/a	n/a	n/a	n/a
Mainstream school/ education setting with a special program for children with hearing loss/ disabilities	n/a	n/a	n/a	n/a	n/a
School/education program for children with hearing loss/ disabilities	n/a	n/a	n/a	n/a	n/a
P value					

HUI3 indicates the Health Utility Index Mark 3; IQR, interquartile range; LL, low language; PedsQL, Pediatric Quality of Life Inventory; SD, standard deviation.
*Significant *P* values are shown in bold.

PedsQL summary score was considered as the minimal clinically meaningful difference.[17]

Statistical Analysis

Because the distributions of the HUI3 and PedsQL scores were skewed on the Shapiro-Francia test[21] ($P < .05$), nonparametric statistical tests of differences were conducted (Wilcoxon-Mann Whitney and Kruskal-Wallis tests). Convergent validity captures whether scores on one instrument correlate with scores on the other instrument designed to assess the same construct.[22] The convergent validity of the PedsQL and HUI3 scores was examined using scatter plots and an assessment of the level of association (Spearman's correlation) between each of the domains of the 2 instruments. Correlations between 0.3 and 0.5 were considered moderate and ≥0.5 strong.[23] We estimated the intraclass correlation coefficients at an individual level based on a 2-way mixed-effect model, where the individual effect was random and the effect of the instrument was fixed to assess the level of agreement between the instruments. An intraclass correlation coefficient less than 0.75 implies poor to moderate agreement, whereas a coefficient greater than 0.75 indicates good agreement.[24] We used Bland-Altman plots to visually examine agreement. For comparability, the PedsQL overall scores were converted to a 0-to-1 scale by dividing the overall scores by 100. Then mean HRQoL overall scores and the difference in HRQoL overall scores between instruments were calculated and plotted against each other. A line of mean difference estimates systematic difference between the two instruments, with limits of agreement estimated as the mean difference plus/minus 1.96 standard deviation of the mean difference. Limits of agreement reflect the expected range in which 95% of observed differences would lie, with wider limits of agreement indicating poorer agreement.[25] Analyses were conducted using Stata version 13.0[26] and MedCalc version 17.9.4 for the Bland-Altman plots (MedCalc software bvba, Ostend, Belgium).

Results

Participant Characteristics

Of the 1012 children in the general population sample, more than half (54%) were male, 12.5% had LL, and almost all parents (94%) had completed secondary school (year 12 in Australia). In the sample of children with LL (n = 126), 67% were male, the mean (SD) age was 4.2 (0.1) years, and again, almost all parents (91%) had completed secondary school. In the sample of children with CHL (n = 108), fewer than half (45%) were males, the mean (SD) age was 5.3 (0.8) years, 40% had LL, and 61% of the parents had completed secondary school.

HRQoL in Children With LL and CHL

In the general population, children with LL had a 6% significantly lower HRQoL (measured by the HUI3) than their peers with typical language. Among the children with CHL, LL was associated

Table 1. Continued

Characteristic	n (%)	Children with congenital hearing loss (SCOUT, n = 108)			
		HUI3		PedsQL	
		Mean (SD)	Median (IQR)	Mean (SD)	Median (IQR)
Whole sample	108 (100)	0.68 (0.26)	0.74 (0.58-0.85)	75.13 (17.05)	78.33 (65.22-87.77)
Hearing loss severity					
Normal (≤25)	10 (9.26)	0.76 (0.25)	0.82 (0.58-1)	72.11 (15.82)	78.8 (54.54-81.82)
Mild	22 (20.37)	0.73 (0.25)	0.85 (0.63-0.85)	77.40 (18.30)	78.26 (71.74-90.22)
Moderate	27 (25.00)	0.76 (0.16)	0.76 (0.65-0.85)	74.94 (19.84)	76.67 (60.87-93.48)
Severe (61-80)	13 (12.04)	0.53 (0.31)	0.62 (0.49-0.71)	72.46 (19.05)	77.17 (65.22-85.87)
Profound (81+)	36 (33.33)	0.61 (0.29)	0.68 (0.53-0.82)	75.71 (14.06)	80.43 (65.76-85.87)
P value		**.008**		.88	
Language ability					
Typical	58 (53.70)	0.79 (0.16)	0.85 (0.73-0.85)	77.18 (16.95)	78.88 (71.74-89.13)
LL	43 (39.81)	0.60 (0.24)	0.62 (0.53-0.75)	72.32 (17.08)	76.09 (61.96-88.04)
P value		<**.001**		.15	
Mainstream school/ education program	66 (61.11)	0.69 (0.25)	0.75 (0.58-0.85)	78.95 (15.97)	81.52 (72.82-90.22)
Mainstream school/ education setting with a special program for children with hearing loss/ disabilities	10 (9.26)	0.68 (0.19)	0.70 (0.63-0.85)	63.49 (11.92)	64.67 (53.26-71.74)
School/education program for children with hearing loss/ disabilities	27 (25.00)	0.64 (0.31)	0.72 (0.58-0.85)	71.64 (18.24)	79.35 (55.43-84.78)
P value		.76		**.003**	

with a 19% lower HRQoL than children with CHL but typical language and 30% lower HRQoL than children in the general population, as measured by the HUI3 (Tables 1 and 2). The PedsQL did not show statistically significant HRQoL differences between children with and without LL in either cohort.

Children with CHL had significantly lower HRQoL than children in the general population, for example, HUI3 mean (SD) scores of 0.68 (0.26) versus 0.90 (0.13) and PedsQL mean (SD) scores of 75.13 (17.05) versus 85.22 (10.13; Table 2). Among children with CHL, current hearing ability was also meaningful; for example, more severe current hearing loss was associated with lower HRQoL, as measured by the HUI3 (Table 1). HRQoL in children with CHL also differed by school/education setting, with children attending a mainstream school having higher HRQoL than those attending a mainstream school/education program with a special program for children with hearing loss or school/education program for children with hearing loss/disabilities, as measured by the PedsQL (Table 1). Furthermore, children with CHL had lower HRQoL than children with LL (HUI3, $P < .001$; PedsQL, $P < .001$). This was true even if the children with CHL had no current hearing or language problem (HUI3, $P < .001$; PedsQL, $P = .01$).

In terms of HRQoL domains, children with LL in the general population had impaired school functioning (PedsQL) and impairment of speech and cognition (HUI3) but no significant impairment of their physical, emotional, and social functioning (PedsQL) or their vison, hearing, cognition, pain, dexterity, or ambulation domains (HUI3; Table 2). Children with CHL had significantly lower physical, social, and school functioning (PedsQL) and lower vision, hearing, speech, cognition, dexterity, and ambulation states (HUI3) than children in the general population. Emotional functioning was not impaired in children with either LL or CHL (HUI3, PedsQL).

Comparison of HRQoL Measured by the HUI3 and PedsQL

Moderate correlation between HUI3 and PedsQL overall scores was evident in the full general population sample ($r = 0.3$, $P < .001$) and in children with LL ($r = 0.3$, $P = .003$) but not in children with CHL ($r = 0.03$, $P = .78$; Supplementary Fig. 1 found at https://doi.org/10.1016/j.jval.2019.07.019). Furthermore, there were low correlations ($r < 0.3$) between each of the HUI3 and the PedsQL domains in the general population and in the groups of children with LL or CHL (Supplementary Table 1).

Intraclass correlations between the 2 instruments were less than 0.75 across all 3 samples (0.39 in the general population, 0.03 in children with LL, and 0.42 in children with CHL). On the Bland-Altman plots, the 95% limit of agreements ranged from –0.23 to 0.33 points when the HUI3 and the PedsQL were used in the general population (Fig. 1). When the 2 instruments were used in children with LL and CHL, the 95% limits of agreements were even wider (ranging from –0.33 to 0.36 in children with LL and from –0.69 to 0.54 in children with CHL). Thus, in all 3 groups, the 95% agreement differences were far wider than the clinically meaningful difference specified for the HUI3 (0.03 points) and the PedsQL (0.045 points, when converted to a 0-1 scale). Notably, in

Table 2. Comparison of health-related quality-of-life domain scores between children with and without low language in the Language for Learning (L4L) general population and between children with congenital hearing loss in the Statewide Comparison of Outcomes (SCOUT) study and children in the L4L general population.*

	Children with typical language in the community-based sample		Children with low language in the community-based sample		*P* value	Children in the community-based sample		Children with congenital hearing loss		*P* value
	n	Mean (SD)	n	Mean (SD)		n	Mean (SD)	n	Mean (SD)	
PedsQL										
Overall	886	85.47 (9.74)	126	83.49 (12.46)	.25	1012	85.22 (10.13)	108	75.13 (17.05)	<**.001**
Physical	886	89.62 (11.50)	126	87.55 (16.29)	.71	1012	89.36 (12.22)	106	78.08 (23.51)	<**.001**
Emotional	886	73.79 (14.89)	126	73.53 (15.91)	1.00	1012	73.76 (15.01)	108	75.37 (15.67)	.17
Social	886	88.05 (12.58)	126	86.48 (14.14)	.47	1012	87.85 (12.78)	108	74.32 (21.32)	<**.001**
School	869	89.57 (12.80)	118	84.60 (16.44)	**.006**	987	88.98 (13.37)	104	71.08 (19.29)	<**.001**
HUI3										
Overall	886	0.91 (0.13)	126	0.85 (0.15)	<**.001**	1012	0.90 (0.13)	108	0.68 (0.26)	<**.001**
Vision	886	0.99 (0.04)	126	0.99 (0.01)	.49	1012	0.99 (0.04)	108	0.98 (0.08)	<**.001**
Hearing	886	0.99 (0.66)	126	0.99 (0.65)	.57	1012	0.99 (0.07)	108	0.73 (0.22)	<**.001**
Speech	886	0.94 (0.11)	126	0.88 (0.14)	<**.001**	1012	0.93 (0.11)	108	0.79 (0.25)	<**.001**
Emotional	886	0.99 (0.04)	126	0.981 (0.05)	.16	1012	0.99 (0.04)	108	0.99 (0.04)	.50
Cognition	886	0.97 (0.08)	126	0.93 (0.11)	<**.001**	1012	0.96 (0.84)	108	0.89 (0.17)	<**.001**
Pain	886	0.96 (0.08)	126	0.95 (0.10)	.82	1012	0.96 (0.09)	108	0.98 (0.05)	**.05**
Dexterity	886	0.99 (0.04)	126	0.995 (0.05)	.45	1012	0.99 (0.04)	108	0.96 (0.18)	<**.001**
Ambulation	886	0.99 (0.02)	126	0.997 (0.02)	.47	1012	0.99 (0.02)	108	0.96 (0.16)	<**.001**

HUI3 indicates Health Utility Index Mark 3; PedsQL, Pediatric Quality of Life Inventory; SD, standard deviation.
*Significant *P* values are shown in bold.

all 3 groups, the Bland-Altman plots showed a funneling effect with stronger agreement as the mean overall score approached 1.0.

Ceiling effects indicating "full health" were observed for 40%, 18%, and 8% of the general population, LL, and CHL groups, respectively. Overall, 90% to 98% of the children scored "full health" in the HUI3 domains of vision, ambulation, and dexterity (all three groups); hearing (general and LL groups); and emotions and cognition (general population) (Supplementary Table 2). In none of the 3 groups was the full range of response alternatives used for all HUI3 items (eg, feeling sad/blue in emotional functioning).

The ceiling effects for the PedsQL overall scores were 3%, 4%, and 2% in the general, LL, and CHL groups, respectively. Correspondingly, in the general and LL groups, only 2 items (walking and running) were scored at the ceiling (no problems) by more than 90% of the children (Supplementary Table 3). No PedsQL item showed a ceiling effect in the CHL group, and in all 3 groups, almost all response alternatives were used in the great majority of the items.

Discussion

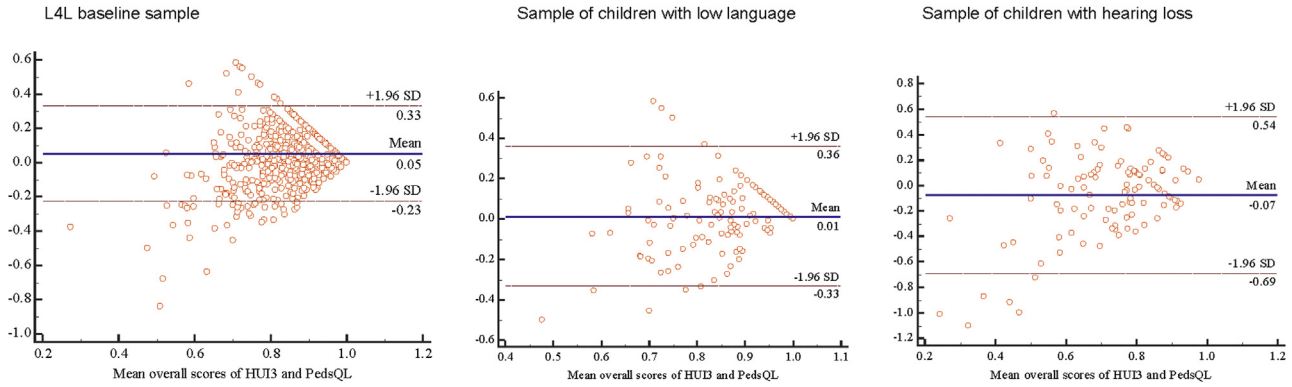
This study investigated HRQoL in children with LL and CHL and in the general population. It is, to our knowledge, the first study to examine the value of concurrently using the HUI3 and the PedsQL in these populations. We found that young children with LL or CHL had significantly impaired HRQoL, with a particularly low HRQoL in children with CHL. In children with LL, the lower HRQoL was due mainly to impairment of school functioning, speech, and cognition. Children with CHL additionally had impaired physical and social functioning, vision, hearing, dexterity, and ambulation. We found low to moderate correlation and low agreement between the HUI3 and the PedsQL across all 3 groups. There was little overlap in the information provided by the specific domains in the 2 instruments, and ceiling effects were high in the HUI3 but not in the PedsQL.

Our HUI3 and PedsQL scores for the 4-year-old Australian children in the L4L cohort (mean scores 0.90 and 85.22, respectively) were comparable to the healthy population norms of younger children in the United Kingdom (mean HUI3 scores 0.92)[27] and in the United States (mean PedsQL score 87.37).[28]

The HRQoL impairment in children with LL reported in our study is consistent with the findings of a previous systematic review concluding that children with speech and language difficulties generally seem to have reduced HRQoL.[3] Our results extend this conclusion to younger children in the preschool years. The review highlights that children with speech and language difficulties have particular problems in social, emotional, and school-related aspects of HRQoL. In our cohort of preschool children, we confirmed impaired school functioning but not social or emotional impairment. This is supported by a recent longitudinal study following HRQoL in children with LL, which showed a shift from impairment of school functioning only at 4 years of age to a more general impairment affecting also social, emotional, and physical functioning at 9 years of age.[29]

Regarding children with hearing problems, a previous systematic review found no HRQoL impairment in children greater than 6 years of age with mild hearing problems[30] but significant impairment in those with permanent CHL.[27] The result of the latter study is consistent with our findings in younger children who primarily had moderate to profound hearing loss. Thus, in younger and older children, more profound hearing loss seems to be associated with impaired HRQoL. The review also showed that hearing loss did not affect physical or social aspects of HRQoL.[4] In contrast, our study found impaired physical and social functioning in children with CHL. Potentially, children in our CHL sample had other medical conditions or intellectual disabilities that affected their physical and social function.

Figure 1. Bland-Altman plot of differences in Health Utilities Index Mark 3 (HUI3) and Pediatric Quality of Life Inventory (PedsQL) scores.



PedsQL total scores were rescaled to 0 and 1 to be comparable with the HUI3 utility scores.

We also found that children with CHL had lower HRQoL than children with LL and that this difference was clinically meaningful. For example, using the preference-based instrument, children with LL might be willing to give up 6% of their remaining life expectancy in exchange for living the remaining life without having LL, whereas the corresponding number in children with CHL was 22%. This difference could partly reflect the exclusion of children with major medical conditions, known intellectual disability, and autism spectrum disorder from the LL but not the CHL sample. Children with both CHL and LL fared worst of all: a 19% reduction in HRQoL compared with children with CHL but typical language (SCOUT) and a 30% reduction compared with children in the general L4L population. We do not know if the different language measures used in SCOUT (PLS4) and L4L (CELF-P2) may have contributed to these differences, but both are widely used and standardized, and the LL definitions were the same.

Regarding the agreement between the 2 instruments, all Bland-Altman plots indicated that the limits of agreement between the HUI3 and the PedsQL were wider than what is considered a clinically meaningful HRQoL difference. Similarly, correlations between subdomains of the HUI3 and the PedsQL were low. Thus, the 2 measures do not seem to provide interchangeable information but rather to supplement each other with unique information.

The HUI3 and PedsQL also showed marked differences in ceiling effects, which were much greater for the HUI3 than for the PedsQL in all 3 groups. The PedsQL may, therefore, be more sensitive to change than the HUI3 in populations of young children. The HUI3 was primarily developed for use in adult populations who were born very premature. Nevertheless, it has been adapted for use in general populations and is also considered applicable for children.[31] Nevertheless, items that are relevant to adults may not necessarily capture core aspects of children's HRQoL. The high percentage reporting ceiling scores in several HUI3 domains (eg, vision and dexterity) suggests that these domains may not be relevant to children in the general population, including those with language or hearing problems.

Strengths of the study include the large community-based sample. We also examined the performance of the 2 instruments in 2 specific cohorts of children with LL or CHL. We used the Bland-Altman approach to explore the limits of agreement between the 2 instruments, which enabled visual examination of agreement across the range of HRQoL that other methods such as Pearson's correlation could not capture.[25]

Study limitations include the use of parent proxy report of HRQoL for both instruments. According to a systematic review comparing parent and child assessments of HRQoL,[32] parents and children are expected to agree on more observable functioning such as physical and motor functioning (eg, ambulation and dexterity) but less on subjective areas such as emotional and social functioning. Because the self-report versions of the HUI3 and the PedsQL are not appropriate for children younger than 5 years, parent proxy report was unavoidable. In addition, the small number of participants in the SCOUT sample limited the reliability of some analyses in relation to children with CHL.

These findings may have clinical implications for population health decision making. For example, although mild CHL may not affect a child's HRQoL, moderate to severe CHL was associated with impaired HRQoL. Thus, interventions or strategies to improve children's health and overall well-being are needed for these 2 groups of children. Furthermore, although less social and emotional impairment in preschool than school-aged children with LL is a good thing, it also presents challenges as to how and when to diagnose and provide intervention for LL. Future research is also needed to validate whether CHL affects children's physical and social function by assessing HRQoL in children with CHL separately for those with and without other medical conditions or intellectual disability.

Our study showed that the PedsQL may not be as sensitive as the HUI3 to distinguish HRQoL impairment in children with and without LL. On the other hand, despite the advantage of being able to produce quality-adjusted life-years, the large ceiling effects in a wide range of the underlying concepts of the HUI3 is problematic because it indicates that this instrument does not solely capture true generic aspects of children's HRQoL. This may limit the ability to detect improvement in HRQoL resulting from interventions targeting young populations. Conversely, including the HUI3 as an HRQoL instrument may be beneficial for studies of HRQoL in children with hearing loss or LL because it has speech and hearing domains that could assist in evaluating the specific effects of hearing loss/LL on children's daily lives.[33]

In summary, young children with LL or CHL had significantly impaired HRQoL, with a particularly low HRQoL in children with CHL. We found poor agreement between the HUI3 and the PedsQL in these child populations. Findings of this study provide insights for future research into HRQoL in young children with these problems. This study also highlights a need for interventions to improve overall health and well-being, targeting children with LL and moderate to severe CHL. The HUI3 and the PedsQL each provide unique information on different aspects of

a child's life and thus can supplement each other in assessing children's HRQoL, especially in pediatric populations with LL or CHL. Nevertheless, high ceiling effects may limit the value of the HUI3 in its general sensitivity to change in populations of young children.

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Supplemental Material

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