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A new tool for the assessment of speech understanding and spatial hearing difficulties in children: the Kid-SSQ questionnaire

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Statements and Declarations

Compliance with Ethical Standard

This clinical prospective study received ethical committee approval (Ile de France VIII, France, N°19.09.02.63029 RIPH3) and was recorded in clinicaltrials.gov (NCT04106063).

Conflicts of Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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ABSTRACT

Purpose - To develop and validate a new questionnaire, the Kid-SSQ, for the rapid screening of hearing abilities in children with hearing impairment, aged 7 to 17 years.

Methods - The questionnaire was constructed from two existing, validated versions of the 'Speech, Spatial and Qualities of Hearing' – (SSQ) questionnaire (pediatric form and adult short-form). The twelve selected items included auditory aspects from three subscales: speech perception, spatial hearing, and qualities of hearing. This new short form was then validated in 154 children with cochlear implants (100 bilateral, and 54 unilateral implanted children). Construct validity was assessed by testing relationships between Kid-SSQ scores and objective clinical parameters (e.g., age at test, pure-tone audiometry-PTA threshold, speech reception threshold-SRT, duration of binaural experience).

Results - Completion time was acceptable for use with children (less than 10 minutes) and the non-response rate was less than 1%. Good internal consistency was obtained (Cronbach's $\alpha = 0.78$), with a stable internal structure corresponding to the 3 intended subscales. External validity showed the specificity of each subscale: speech subscale scores were significantly predicted ($r=0.32$, $p<0.001$) by both 2 kHz PTA threshold ($\beta=0.33$, $p<0.001$) and SRT ($\beta=-0.23$, $p<0.001$). Children with more binaural experience showed significantly higher scores on the spatial subscale than children with less binaural experience ($F(1,98)=5.1$, $p<0.03$) and the qualities of hearing subscale scores significantly depended on both age and SRT ($r=0.32$, $p<0.001$).

Conclusions - The Kid-SSQ questionnaire is a robust and clinically useful questionnaire for self-assessment of difficulties in various auditory domains.

Key words: cochlear implant, quality of life, speech perception, spatial hearing, health questionnaire, hearing loss, hearing aid

List of abbreviations

BCI: bilateral cochlear implant; CI: cochlear implant; PTA: pure tone auditory; SRT: Speech Reception Threshold; SSQ: speech spatial and qualities of hearing; UCI: unilateral cochlear implant

INTRODUCTION

The expansion of clinical indications for cochlear implantation in recent decades (e.g. [2]) has greatly improved treatment for severe to profound bilateral deafness in children. While this has notably improved these children's language development (for review, see [3]), many cochlear implant (CI) children still complain of difficulties in sound localization and speech understanding in noisy environments, even several years after implantation. These difficulties are largely undiagnosed and unreported during clinical follow-up, and can impair long-term social integration, making their detection and clinical care an important public health issue. Thanks to their ease of use and sensitivity, quality of life questionnaires are commonly-used screening tools. However, most auditory quality of life questionnaires consider disability as a whole, and lack specificity for the diagnosis of auditory difficulties (see for example the KINDL Cochlear Implant Module questionnaire adapted for young CI users[4]). Designed for adult population with hearing-impairment, the Speech, Spatial, and Qualities of Hearing Scale (SSQ)[5], is aimed at encompassing the widest range of aspects of hearing in daily life of patients and, with its availability in multiple languages and its various forms, has been used in more than 180 international publications, showing its usefulness and relevance in the audiological field. This 49 item long scale consists in depicting a situation met in daily life (e.g., *"You are in a group of about five people in a busy restaurant. You can see everyone else in the group. Can you follow the conversation?*"), and inviting patients to answer on a 11 points visuo analog scale, graded from 0 ("*Not at all*") to 10 ("*Perfectly*"). Patients have the option to tick a "not applicable" (NA) box if they feel the situation depicted doesn't apply to them. The 49 items are organized in 3 subscales (i.e. *speech perception*, *spatial hearing*, and *qualities of hearing*), that have shown to provide, each, a specific index of hearing abilities[6, 7]. Unfortunately, the wealth of information brought by this more than 1000 word long scale comes at the price of a cognitive burden for the patients[8], that can compromise its acceptability, and its feasibility in clinical settings. To overcome this problem, several short forms have been built[9], the latest, in 15 items, (15iSSQ[10]) being created based on a data-driven approach, showing good psychometric properties and keeping the 3 original subscales.

Adapting adult scales for pediatric population involves adapting language, response types and length of scale[11–13]. Galvin and Noble [14] adapted the original adult SSQ for the assessment of children, taking specific care in adapting the wording and the situations depicted to children's daily lifes. While it explores a broad range of everyday auditory skills, through the same 3 subscales as the original SSQ, this pediatric adaptation, aimed at children above 10 years of age, consists of 33 items and takes approximately 30 minutes to complete. Responses are therefore subject to bias due to attentional difficulties, which are particularly prevalent in young children with hearing loss.

Moreover, some items, in the long SSQ versions, have a high non-response rate due to the complexity of the everyday situations they describe [7, 15, 16]. Length of scale is part of the response burden of a scale[12], and should be optimized so that it doesn't compromise the acceptability, nor reliability of the scale, hence our choice of aiming for a 12 item long scale, allowing 4 items per subscale, with specific adaptation for children.

The primary objective of this study was thus to develop and validate a short version of the SSQ for children, based on Galvin and Noble's work [14], and using the same data-driven approach as that used to develop the 15iSSQ short form [10], that is in agreement with the international guidelines regarding translation and cultural adaptation of health questionnaires[17, 18], validation of health questionnaires[19] and creating short-forms[20, 21].

The second objective was to assess the construct validity of this new short-form, in particular how this scale relates to several clinical variables in children fitted with one or two CIs, in order to propose a new routine assessment tool for subjective hearing difficulties encountered in daily life.

METHODS

Participants

One hundred bilateral cochlear implant children (BCI) aged 7.3 to 17.8 years (mean ± standard deviation; 12.5 ± 2.9 years) and 54 unilateral cochlear implant children (UCI) aged 7.7 to 17.8 years (13.8 ± 2.2 years) were included in this study. Four children in the UCI group had bilateral implants before inclusion in the study but had opted to maintain only one CI, and 67% of UCI children had a contralateral hearing aid. For the BCI and UCI groups the three main hearing loss etiologies were "unknown" (42% and 39%), "genetic" (35% and 33%), and "acquired" (9% and 11%). In terms of schooling, 21% of the BCI children and 39% of the UCI children were enrolled in adapted schooling. The demographic and audiometric characteristics of each group are summarized in online resource, Appendix 1.

Questionnaire construction

We used two versions of the Speech Spatial and Qualities of hearing (SSQ) questionnaire as our reference: its short form for adult hearing assessment (the 15iSSQ [10]) and its long form for children (the SSQ33 [14]). Based on these two questionnaires, we developed a short pediatric form which we named the Kid-SSQ. A short questionnaire created from one or more forms of a validated questionnaire should [21] : 1) maintain the same internal structure as the reference forms, i.e. same number of subscales, 2) include between 3 and 5 items per subscale [22], and 3) exclude items with poor psychometric properties (e.g., high non response rates) and poor discriminability between pathological condition and controls. As the 15iSSQ was built using this data-driven approach, encompassing data and results from several international studies, we used the same criteria to create the Kid-SSQ. In addition, we also used data available from the long form for children SSQ [14] and our clinical experience of its use [15]. We included 4 items from each of the 3 subscales of the already-validated versions of the SSQ questionnaire, each representing a different hearing function (subscale A: speech perception, subscale B: spatial hearing, subscale C: qualities of hearing). Each item was assigned a letter (subscale) and a number (item) (e.g. for subscale A, each item was labelled from #A.1 to #A.4, for subscale B from #B.1 to #B.4, and for subscale C from #C.1 to #C.4. Items were selected based on their psychometric properties, on their performance in the SSQ33, and with the goals of covering a wide range of hearing situations [23] and being understood by children as young as 7 years old. The Kid-SSQ shares 9 of its 12 items with the 15iSSQ, and all 12 items are in the long pediatric version of the SSQ. An abbreviated text of the communication situation depicted in each of the items is provided in Table 1 and details of the items included in the Kid-SSQ can be found in the Online Resource, Appendices 2 and 3.

We used the validated, French language version of the SSQ [8] to create a new, pediatric short-form. We retained the same rating scale as that used in the two reference forms: zero for "I can't do it at all" to ten for "I can do it perfectly". To assist younger children with the self-assessment task we added three smileys ("sad" for a rating of 0", “neutral” for a rating of 5, and "happy" for a rating of 10), as well as a color gradient (light for 0, to dark for 10). We also modified the shape of the scale to look like a funnel mimicking the increasing intensity of the response (see online resource, Appendix 4A). Despite these additions, pilot experiments revealed that a minority of children under 9 were still unable to use the numerical scale, so an additional scale was developed without numbers, based on the face scale used for pain assessment (Appendix 4B). The color scheme and the chosen smileys are widely used in French schools from nursery level onwards. Those modifications are among those typically recommended by international guidelines for pediatric health scales creation[11, 13]. The original questionnaire in French is available in online resource, Appendix 5. The questionnaire was administered only once, using a paper and pen format during a face-to-face interview with each child. The items were read aloud by the experimenter (or by the child) and after reading the item, if necessary, the experimenter provided an example from everyday life so that the child could more easily imagine him/herself in the described situation. Children were asked to choose a whole number and were told they could choose the response "not applicable" (NA) if they were unable to answer the item.

Subscales	**Item number**	**Kid-SSQ**	**Item number**
	Kid-SSQ	**Listening situations (abbreviated text)**	**SSQ33**
Speech perception	**A1**	Talking with one person with TV on	**1.1**
	A2	Conversation with 5 people in noise with vision	**1.4**
	A3	Conversation with 5 people in noise without vision	**1.6**
	A4	Follow one conversation when many people talking	**1.8**
Spatial hearing	**B1**	Locate a door slam in a familiar house*	**2.4**
	B2	Locate dog barking	**2.5**
	B3	Locate vehicle from footpath	**2.6**
	B4	Identify approach or recede (voice or footsteps)	**2.8**
Qualities of hearing	**C1**	Identify different people by voice	**3.3**
	C2	Distinguish familiar music	**3.4**
	C3	Distinguish 2 different sounds (running water /fan)	**3.5**
	C4	Recognize different familiar sounds in the house	**3.2**

Table 1 *List of the listening situations depicted in the 12 items of the Kid-SSQ (the text of each item has been abbreviated, adapted from Bahn et al. (2012*[24]*), and not the actual text presented to children). The correspondence of the item numbers of the Kid-SSQ with the item numbers of the original pediatric scale (SSQ33) are mentioned in the rightmost column. (Further correspondence between the different forms of the SSQ scale are in the online resource Appendix 2).*

Statistical analyses

The validation procedure involved the different steps recommended for statistical validation of health scales [19]. Internal consistency of the Kid-SSQ was assessed by combining data from all patients and calculating both item-to-total correlations and Cronbach's alpha for each factor determined by factor and cluster analyses. Exploratory factor analysis was performed using the same parameters as previously used for the SSQ [6, 8, 10], namely a factor extraction performed on a non-parametric correlation matrix with a maximum likelihood method and an oblique rotation method. The details of this analysis is in the online resource, Appendix 6.

The external validity of the Kid-SSQ was evaluated by Pearson correlation and multi-regression analysis, between the mean score per subscale and various clinical parameters (i.e. age at test, pure tone auditory threshold - PTA, speech reception threshold - SRT, duration of exposure with the first CI, duration of binaural experience, and inter-implant interval).

RESULTS

Kid-SSQ responses

The number of non-responses or "not applicable" (NA) answers was very low (<1%). Most of the "NA" answers were distributed between two items: 5 for #C.2 *(Identify different people by voice)* and 4 for #B.4 *(Identify approach or recede)*. Only 12 children had missing answers and 11 of these missed only one answer (one child missed 2). The median age of children with and without missing answers was similar (13 years). The face scale (Appendix 4B) was rarely necessary, only 5 BCI children under 9 years old used it.

The average total score on the Kid-SSQ for the whole sample was 6.6 (±1.3) out of 10, and did not differ between the BCI and UCI groups (see ANOVA results below). Figure 1 and online resource Appendix 7 shows the scores for each subscale and item separately for the BCI and UCI groups. A repeated measures ANOVA revealed that subscale scores differed between each other ($F(2, 304) = 131.86$, $p < .001$, $\eta_g^2 = .27$), but that scores on the three subscales were similar for both groups (no main effect of group nor a group by subscale interaction). Post-hoc tests revealed that subscale C scores were higher than scores on the other two subscales (p<.0001, Bonferroni correction). Since the UCI group consisted of children with bimodal (i.e. fitted with a CI and a hearing aid) and monaural (i.e., with only a CI) configurations, scores for these two sub-groups are shown in Appendix 8. Monaural UCI children tended to have lower scores on all three subscales.

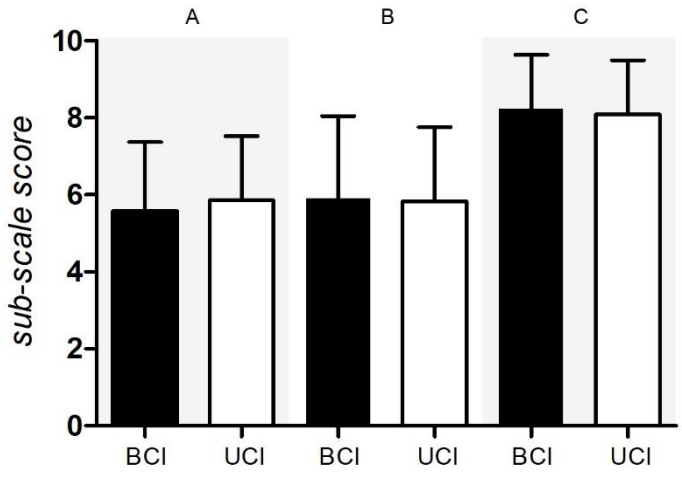

Fig. 1 *Means and standard deviations on the three subscales for the bilateral cochlear implant (BCI, n=100) and unilateral cochlear implant (UCI, n=54) children (A: speech perception; B: spatial hearing; C: qualities of hearing)*

Factor analyses and internal consistency and reliability

The item to total correlations ranged from 0.22 (#C2) to 0.53 (average of 0.42), showing good internal consistency and Cronbach's alpha was 0.78 (0.76; 0.80) for the entire scale, and remained above 0.75 as each item was removed (Appendix 9).

The common factor analysis performed on 142 children explained 42% of the total variance with three clearly defined factors, and average communalities across items of 0.42, all communalities were above 0.21 (#C2) (factor loadings and communalities are detailed in Appendix 9). #A2 *(Conversation in noise with vision)* and #A3 *(Conversation in noise without vision)* showed a clear load on factor 1 (16% variance explained), with #A1 (*Talking with one person with TV on*) & #A4 (*Follow one conversation when many people talking*) cross-loading on factor 1 and 3 (#A4 loading more on factor 3). All B items loaded on factor 2 (13% of variance explained), with #B3 (*Locate vehicle from footpath*) and #B4 (*Identify approach or recede*) cross-loading on factors 2 and 3. #C2 *(Identify familiar music)*, #C3 *(Separation of two sounds*) and #C4 *(recognize different familiar sounds)* clearly loaded on factor 3 (14% variance explained), with #C1 *(Identify different people by voice)* cross-loading on factors 2 and 3. This internal structure was confirmed by the cluster analysis (Figure 2), which identified 3 higher-order sub-clusters with good internal consistency and homogeneity, each comprising 4 items from the corresponding subscales of the questionnaire (cluster C10: #A1 to #A4; cluster C7: #B1 to #B4 and cluster C8: #C1 to #C4). Attempts to link these higher-order clusters together (i.e. C9, C10, and C11) resulted in a loss of homogeneity, evident in the larger differences between alpha and beta coefficients. This shows that our 3 higher-order clusters could not be combined into a single score, suggesting that the scale is not unidimensional, but instead explores 3 different aspects of hearing in our population. Correlations between the 3 different subscales ranged from 0.33, $p<5x10^{-5}$ (A and C scales) to 0.41, $p<3x10^{-7}$ (between B and C scales).

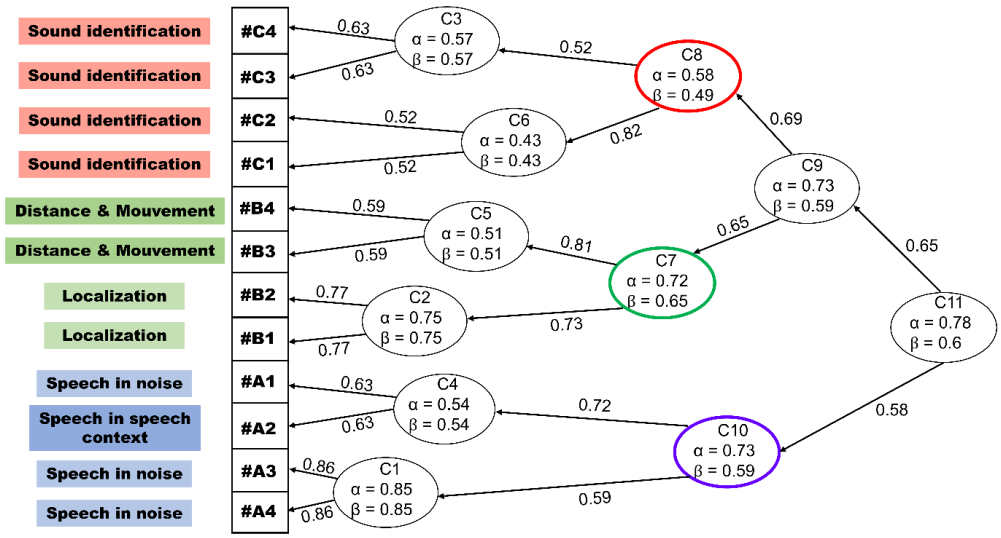

Fig. 2 *Tree diagram of the cluster analysis. The 12 questionnaire items (4 from each subscale) are shown on the left in the colored rectangles and the most similar items are combined on the left of the figure. The three main clusters are circled in red, green, and blue, and correspond to the C, B, and A subscales of the questionnaire. For each cluster, the alpha and beta coefficients are indicated.*

External validity – correlation with clinical parameters

For the BCI group, the Kid-SSQ total score did not correlate significantly with CI experience, PTA threshold, or SRT. A weak but significant increase in scores was observed with increasing age (r=0.23, p=0.02), mainly due to the significant correlation between age and the subscale C score *(qualities of hearing)* (r=0.31, p<0.002), due to correlations between age and #C3 (r=0.26, p=0.001) and #C4 (r=0.23, p=0.004). Higher Kid-SSQ total scores were associated with higher binaural (averaged across right and left ears) PTA thresholds (r=0.19, p=0.055) and higher binaural 2 kHz PTA thresholds (r=0.18, p=0.06). Although those trends were not statistically significant, they can be attributed to the high correlation between subscale A scores and binaural PTA thresholds (r=0.29, p<0.003), and subscale A scores and binaural 2 kHz PTA threshold (r=0.28, p<0.005).

The different subscales showed different correlation patterns. First, subscale A *(speech)* scores correlated significantly with PTA threshold averaged over 0.5 to 4 kHz on both ears in the BCI group (r=0.29, p=0.003): scores improved as the average threshold increased. Similar results were obtained between speech scores and both left ear (r=0.25, p=0.012) and right ear hearing thresholds (r=0.25, p=0.012).

	Total Score	A Speech score	B Spatial score	C Qualities score	SRT	PTA	2 kHz PTA
Speech score	**0.75*** **						
Spatial score	**0.82*** **	**0.38*** **					
Qualities score	**0.7*** **	**0.33*** **	**0.41*** **				
SRT	-0.13	-0.1	-0.07	-0.15			
PTA	0.04	0.15	-0.03	-0.04	**0.48*** **		
2 kHz PTA	0.15	**0.24* **	0.05	0.04	**0.38*** **	**0.85*** **	
Age	**0.22* **	**0.17**	0.1	**0.27** **	0.04	0.1	0.12

Table 2 *Pearson correlation coefficients between the different Kid-SSQ subscales and main auditory variables, in a population of 149 cochlear implant children (listwise deletion from the total population). Values in bold are statistically significant (p<0.05), with * for p<0.01, ** for p<0.001 and *** for p<0.0001.*

When considering the first implanted ear for the entire population (Table 2), the subscale A score tended to increase as the PTA increased (r=0.15, p<0.07). More detailed analyses revealed that this correlation was significantly stronger for the 2 kHz hearing threshold (HT) (r=0.24, p<0.01) than with for any lower frequency (e.g. r=0.09 for 1 kHz HT, p=ns, z=2.25, p<0.025) and that among the A items, A3 scores increased significantly with decreasing SRT thresholds (r=-0.22 p=0.008). The best explanatory model for A scores (F(2, 148)=8.5, p<0.0004, r=0.32) involved both SRT and 2 kHz HT as significant predictors, with 2 kHz HT being the strongest predictor (β =0.33, t=3.9, p<0.0002) versus SRT (β =-0.23, t=-2.7, p=0.007). When controlling for the 2 kHz HT, the A score decreased significantly (by 0.5 points) as SRT increased by 10 dB. Similar significant models were obtained for both A2 and A4, but they were not statistically significant for A1. For A3, SRT was the major predictor (β =-0.32) versus the 2 kHz HT (β=0.27), with a 0.94 points decrease in A3 score for each 10 dB increase in SRT.

Subscale B (spatial) scores tended to increase with longer experience with the second CI (r=0.173, p=0.086), and were not significantly correlated with age (r=0.11, p=ns) or experience with the first CI (r=0.03, p=ns), or ear asymmetry (r=0.15, p=ns).

When the group of BCI patients was median split based on amount of experience with bilateral CI, the Kid-SSQ total scores tended to be higher in the group with more bilateral CI experience (F(1,98)=3.9, p=0.05), and the subscale B (spatial) score was significantly higher in this group (F(1,98)=5.1, p=0.026). This was particularly true for B1 and B4.

Subscale C (qualities of hearing) scores increased significantly as age increased ($r=0.29$ $p<0.0004$), and with amount of experience with a first cochlear implant ($r=0.17$, $p<0.04$), but this latter effect was entirely mediated by age (direct effect: $z=0.5$, p=ns, indirect effect via age: $z=2.6$, $p<0.01$). The best predictive model for C scores ($F(2,148)=8.1$, $p<0.0005$, $r=0.32$) involved both age ($\beta=0.28$, $t=3.5$, $p<0.0007$) and, to a lesser degree, SRT ($\beta=-0.16$, $t=-2.1$, $p<0.04$) with a decrease in C score of 0.3 points as SRT increased by 10 dB, after having controlled for age. Detailed analysis of the subcluster formed by C1 *(Identify different people by voice)* and C2 (*distinguish familiar music*) showed a strong prediction only by SRT ($r=0.23$, $p=0.005$), with a decrease of 0.47 (se=0.16) by 10 dB increase in SRT. The subcluster formed by C3C4 showed a significant increase of 0.2 points (se=0.05) per year of age ($r=0.30$, $p<0.0002$).

DISCUSSION

Assessment of quality of life in CI children is an essential part of their medical follow-up and clinical care. It is complementary to objective audiological testing and allows early detection of persistent hearing problems. In this study we developed a new short form of the pediatric SSQ, called the Kid-SSQ, and we validated it in a large cohort of 154 CI children. This new short form has 5 strengths: (1) it is specific to auditory disorders, (2) it is adapted for use with a large range of ages: from 7 to 17 years old, (3) completion is fast and simple, making it possible to perform during a medical visit, (4) it is sensitive enough to accurately quantify children's complaints, and (5) it has the same internal structure of 3 different subscales as the original SSQ, with each subscale being specific to a different aspect of hearing. In the literature, we did not find similar scales in other languages adapted in a short format for use in clinical practice.

Validation of a new short-form of the pediatric SSQ

The Kid-SSQ questionnaire consists of twelve items (3 subscales with 4 items each), selected from the original scale developed by Galvin and Noble (2013), using the same data-driven approach for short-form development as that used in developing the adult 15iSSQ short-form [10]. These items explore a wide variety of auditory functions: understanding speech in noise, spatial hearing of a stationary or moving source, identification of everyday sounds. Similar to both the original pediatric and adult scales, it has an internal structure with three well defined subscales (A- speech perception, B- spatial hearing, C- qualities of hearing), obtained by exploratory factor analysis and confirmed by cluster analysis. In our sample it achieved a Cronbach's α of 0.78, showing good reliability for a 12 item scale (as Cronbach's alpha decreases with the number of items [25]). The consistency

indices revealed that items correlated well with each other, without multicollinearity, and thus showed a wide range of constructs.

The format and content of this new questionnaire were adapted to make it appropriate for self-assessment by children less than 10 years old. Its completion time is less than 10 minutes and the numerical scale with smileys allowed children to easily rate their difficulties in a variety of daily-life situations (non-response rate was < 1%). In this study, we validated it using more than 150 children, the minimum requirement for stable exploratory factor analysis being 120 children (corresponding to ten children per item) [18, 24].

The multidimensional aspect of this short form is revealed by different response patterns on each subscale (i.e. notably a higher mean score on subscale C-qualities of hearing), by the cluster analysis, and by differences in correlation patterns between subscales and clinical parameters.

Subscale A scores (*speech*) were positively correlated with average PTA, and especially the 2 kHz hearing threshold (and to a lesser degree 4 kHz hearing threshold). While at first glance this result seems counterintuitive, since all items in subscale A depict situations of speech understanding in noisy environments, one possible explanation is that higher PTA at high frequencies leads to reduced perceived noise, an overall improvement in signal-to-noise ratio due to greater filtering of the higher frequencies, and thus better perception in noisy environments. Furthermore, after controlling for the 2 kHz hearing threshold, subscale A scores significantly increased (by 0.5 points) per 10 dB SRT decrease. This is particularly true for the #A3 item, pertaining to hearing in noise without visual clues: its score depends more on SRT than on 2 kHz hearing threshold, increasing by almost 1 point per 10 dB decrease of SRT: this shows the importance of speech perception when relying on purely auditory cues. For items #A2 *(Conversation with 5 people noise with vision)* and #A4 (*Follow one conversation when many people talking*), where visual clues are available and could help, the score is more closely linked to the 2 kHz hearing threshold, i.e., a threshold that allows a greater filtering-out of frequencies above 2 kHz, than to the SRT. This further emphasizes the importance of this type of questionnaire, i.e., evaluating situations encountered in daily life by children with cochlear implants, and the additional need to assess speech perception in environments closer to those frequently met every day (e.g. classroom conversations).

Subscale B (*spatial hearing*) scores increased with amount of binaural experience but did not correlate with inter-implant interval, contrary to the findings of a study using objective evaluation of spatial hearing abilities [15]. It is well known that inter-implant intervals greater than 18 months lead to asymmetric auditory function [27, 28] which could directly impair spatial hearing. However, it is possible that more than 24 months of auditory experience after the second CI could be sufficient

to counteract auditory asymmetry and improve spatial hearing abilities [29]. Here, only #B1 (*Locate a door slam at home)* and #B4 *(Identify approach or recede)* showed significantly greater scores for children with more binaural experience: both these items relate to localization in familiar and commonly-encountered situations (a door slamming at home, sounds made by moving sources such as people voices or steps), whereas #B2 (*Locate dog barking*) and #B3 *(Locate vehicle from footpath)* refer to localization in noisier and less familiar environments (street, outside). One possibility is that the situations depicted in items #B2 and #B3 are more difficult.

Subscale C scores (*Qualities of hearing*) correlated positively with duration of exposure to the first CI and age at test. Mediation analysis showed that the entire effect of first cochlear implant experience was mediated by age. Cluster analysis identified 2 subclusters within scale C. The first one, C1C2 (*Identify different people by voice and distinguish familiar music)* significantly increased its scores (by almost 0.50 points) per 10 dB of SRT decrease, whereas the second one, C3C4 *(distinguish and recognize different sounds)*, was heavily dependent on age. C1C2 deals with recognition of familiar sounds, and implies that this occurs in a quiet environment, which could explain the significant correlation with SRT, and not with PTA (by contrast with the A scores). Conversely, both #C3 and #C4 depended exclusively, and significantly, on age. These two items question the ability to separate and recognize two sounds close to one another, which is a particularly difficult skill. This suggests that this particular auditory function is driven by auditory maturation in childhood but that it also relies on early cochlear implantation, during the critical period. Indeed, many studies have shown that profound disruption of auditory function during the first year of life can jeopardize auditory development [30, 31] and lead to detrimental cortical reorganization. Furthermore, this evolution as a function of age reveals the importance of assessing Kid-SSQ scores across various ages in a normal-hearing population of children. Such data are necessary to ascertain if the strong correlation we observed here with age is specific to children with cochlear implants, or if it is linked to normal auditory neurodevelopment. Indeed, several studies have shown that children as old as 11 have significantly greater difficulty than young adults in perceiving speech in cocktail party noise [32]. Since only C3C4 items showed a strong correlation with age, it is unlikely that this correlation can be explained by difficulties younger children had in understanding the items. Indeed, the qualities SSQ items whose scores correlated with the number of years of education in an elderly population [7] (and therefore were more difficult to understand), have been eliminated from the 15iSSQ, and hence not included in the Kid-SSQ.

Comparison with the reference form

Galvin and Noble [14] were the first to develop a specific pediatric hearing questionnaire – the SSQ33. Initially conceived to evaluate hearing difficulties for all children older than 10 suffering from hearing loss, its use is largely limited to clinical research, because it requires at least 30 minutes to complete. To our knowledge, no French deafness referral centre uses the SSQ33 (or a pediatric equivalent) as a clinical follow-up instrument for children with hearing aids or CI. This means that there is currently a lack of knowledge about the evolution of quality of life experienced by children themselves (and not their parents) over the years after fitting.

Rauch and colleagues [16] recently used the pediatric version of the SSQ33 to evaluate the long-term (3.5 years) outcomes of cochlear implantation in 11 children with profound hearing loss. Ten of the eleven children were between 5 and 9 years old, and only 5 children in the study successfully completed the questionnaire. Three years after cochlear implantation, the average scores of the SSQ33 were greater than what we observed for our 12-item version (subscale A: 8.6; subscale B: 7.3; subscale C: 8.2). This difference is attributable to the less severe hearing loss profile of the children in their study, who all had normal hearing in the ear contralateral to the CI, as well as to the fact that most of the children were less than 10 years old (younger than the lower limit fixed by Galvin and Noble [14]).

We recently evaluated spatial hearing performance in 18 BCI children between 8 and 17 years old using a 3D localization test and the pediatric SSQ33 [15]. No correlation emerged between objective spatial hearing abilities and children's self-assessments. Moreover, mean scores were greater than expected and similar across subscales (A: 7.2; B: 7.1; C: 7.6). It seems that the SSQ33 was not sensitive enough to highlight hearing difficulties in CI children because of its length and the inclusion of several items not relevant to children's understanding or lifestyle (e.g.: "speech in quiet", "sound naturalness"). Self-assessed hearing difficulties measured using the SSQ33 have probably been underestimated because of this scale's inclusion of insensitive items and items with high non-response rates.

Alternative forms to the long form of the pediatric SSQ and future research areas

Galvin and colleagues developed an alternative version of the SSQ for use by parents of children less than 6 years old [14, 33]. This questionnaire was adapted from the long form of the adult SSQ [5] and consists of 23 items. Its administration differs from the SSQ33 for children because the authors recommend that, to obtain accurate responses, parents should observe their child in daily life for 3 weeks (i.e. one week per subscale). In clinical practice, this type of monitoring is restrictive and runs a high risk of poor compliance.

The main use of this SSQ for parents has been in studies that aim to evaluate the benefits of bilateral cochlear implantation [34–37]. In these studies, parents completed the questionnaire before surgery (i.e., when children were in the UCI condition) and up to 2 years after (in the BCI condition). These studies revealed that mean scores for the three subscales increased significantly after 2 years of binaural experience, with the greatest benefits observed on the spatial hearing items. However, in this young pediatric population, scores should be interpreted cautiously because some items can be difficult to score since they do not correspond to daily life of the youngest patients (e.g., "group conversations in a noisy environment" for children under 3 years old, see Lovett [35]). The test/retest reliability of the SSQ for parents and for children has recently been evaluated using a French language version of the pediatric SSQ scales [38], and has shown extremely good reproducibility for both children and parents ($r>0.90$ for each subscale) with test/retest at a two weeks interval. Furthermore, scores from a group of normal hearing children have been found to be above 9 (out of 10) for the majority of items, with a standard deviation around 1.0, showing very good contrast with hearing impaired children. This is extremely encouraging as to the test/retest reproducibility of the Kid-SSQ and its discriminative power. As the majority of the Kid-SSQ items are shared with the 15iSSQ, and as the 15iSSQ has shown better sensitivity and specificity than the original SSQ long form as well as other short-forms [10], the Kid-SSQ should perform well in discriminating between pathological and control conditions. The results of the present study provide new, complementary information about self-perceived speech-in-noise abilities in children with cochlear implants. Speech scores improved by around 0.5 scale points (out of 10) per 10 dB of improvement in SRT. However, contrary to expectation, scores deteriorated with decreasing high frequency hearing threshold, probably because of a greater filtering-out of noise. An unusually bad score in the Speech scale associated with good SRT thresholds could trigger more detailed audiological assessment of speech in noise, to see if speech perception in noise could be improved by minor adjustments in high frequency pure-tone thresholds. Further validation studies are needed to investigate test-retest reliability, to establish sensitivity to changes in auditory skills across time, and to establish age-stratified norms in a normal-hearing population. It would also be interesting to expand this validation to children with a larger variety of auditory disorders, such as mild-to-moderate hearing loss and auditory processing disorders.

Conclusion

The 12-item Kid-SSQ questionnaire, presented and validated here in a large cohort of children with cochlear implants, showed good consistency, very good construct validity and feasibility in children. In addition, this questionnaire shares the same internal structure of the adult SSQ, with 3 subscales, each assessing a different aspect of hearing (speech in noise, spatial abilities and qualities

of hearing). This will permit the assessment and follow-up of three different aspects of hearing. Furthermore, because the questionnaire reflects difficulties encountered in daily life, it provides new and complementary insights into such difficulties to those provided by classic audiological tests. Not only can the Kid-SSQ be used in clinical practice to monitor the benefits of cochlear implantation and speech therapy rehabilitation over time, but it can also be used in children with a large variety of auditory disorders, such as mild-to-moderate hearing loss and auditory processing disorders. This would potentially enable all children with hearing difficulties to benefit from more effective clinical care and follow-up.

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Captions

Fig. 1 *Means and standard deviations on the three subscales for the bilateral cochlear implant (BCI, n=100) and unilateral cochlear implant (UCI, n=54) children (A: speech perception; B: spatial hearing; C: qualities of hearing)*

Fig. 2 *Tree diagram of the cluster analysis. The 12 questionnaire items (4 from each subscale) are shown on the left in the colored rectangles and the most similar items are combined on the left of the figure. The three main clusters are circled in red, green, and blue, and correspond to the C, B, and A subscales of the questionnaire. For each cluster, the alpha and beta coefficients are indicated.*

Table 1 *List of the listening situations depicted in the 12 items of the Kid-SSQ (the text of each item has been abbreviated, adapted from Bahn et al. (2012*[24]*), and not the actual text presented to children). The correspondence of the items numbers with the item number of the original pediatric scale are mentioned in the rightmost column. (Further correspondence between the different forms of the SSQ scale are in the online resource appendix 2).*

Table 2 *Pearson correlation coefficients between the different Kid-SSQ subscales and main auditory variables, in a population of 149 cochlear implant children (listwise deletion from the total population) . Values in bold are statistically significant (p<0.05), with * for p<0.01, ** for p<0.001 and *** for p<0.0001.*

A new tool for the assessment of speech understanding and spatial hearing difficulties in children: the Kid-SSQ questionnaire

Online supplementary material

Appendix 1: Demographic and audiometric data of the 154 cochlear implant children included in the study.

		BCI n=100	**UCI** n=54
Demographic data	Sex ratio (M:F)	58:42	29:25
	Age at diagnosis (months)	17.6 (14.2)	22.2 (19.8)
	Age fitted with hearing aids (months)	19.0 (13.8)	26.5 (19.4)
	Age at CI1 (months)	41.0 (30.0)	62.1 (45.8)
	Inter-implant interval (months)	38.8 (35.3)	NA
	Number of simultaneous CIs	15	NA
	Number of UCI with contralateral HA	NA	36
	Number of left CI1	31	17
	Number of right CI1	54	37
	Age at testing (years)	12.5 (2.9)	13.8 (2.2)
	Conventional schooling	79	33
	Adapted schooling	21	21
Audiometric data	PTA of left ear (dB HL)	25.5 (5.9)	NA
	PTA of right ear (dB HL)	24.4 (5.7)	NA
	PTA of the CI without HA (dB HL)	NA	26.0 (7.0)
	SRT with two CIs (%)	32.7 (6.7)	NA
	SRT with one CI without HA (%)	NA	36.7 (9.7)
	SRT with one CI with contralateral HA (%)	NA	34.4 (6.6)

BCI: bilateral cochlear implant; CI: cochlear implant; CI1: first cochlear implant; HA: hearing aid; PTA: pure tone auditory threshold; UCI: unilateral cochlear implant;
Values are means and standard deviations are in brackets.
One hundred children were included in the BCI group and 54 in the UCI group (which included 36 children with a contralateral hearing aid).
The PTA was calculated for each ear. The speech reception threshold was measured with two CIs for the BCI group (n=100); with one CI only (n=18) or with one CI plus a contralateral HA (n=36) for the UCI group.

Appendix 2: Item comparisons from the original adult form (SSQ), the long pediatric form (SSQ33), the short adult form (15iSSQ) and our proposed short pediatric form (Kid-SSQ).

Subscales	SSQ	SSQ33	15iSSQ short form	Kid-SSQ short form
	Abbreviated text (from Bahn et al., 2012)	**(from Galvin et al., 2013)**	**(from Moulin et al., 2019)**	**(from present study)**
A-Speech perception	Talking with one person with TV on	**x**	**x**	**x**
	Talking with one person in quiet room	**x**		
	Conversation 5 people quiet with vision	**x**		
	Conversation 5 people noise with vision	**x**	**x**	**x**
	Talking with one person in continuous noise	**x**	**x**	
	Conversation 5 people noise without vision	**x**	**x**	**x**
	Having conversation in echoic environment	**x**		
	Ignore interfering voice of same pitch			
	Ignore interfering voice of different pitch			
	Talk with one person and follow TV			
	Follow one conversation when many people talking	**x**	**x**	**x**
	Follow conversation switching in a group	**x**		
	Have conversation on telephone	**x**		
	Follow one person speaking and telephone			
B-Spatial hearing	Locate lawnmower	**x**		
	Locate speaker around a table	**x**	**x**	
	Lateralize a talker to left or right	**x**		
	Locate a door slam in unfamiliar house*	**x**		**x**
	Locate above or below on stairwell			
	Locate dog barking	**x**	**x**	**x**
	Locate vehicle from footpath	**x**	**x**	**x**
	Judge distance from footsteps or voice	**x**		
	Judge distance of vehicle	**x**		
	Identify lateral movement of vehicle	**x**		

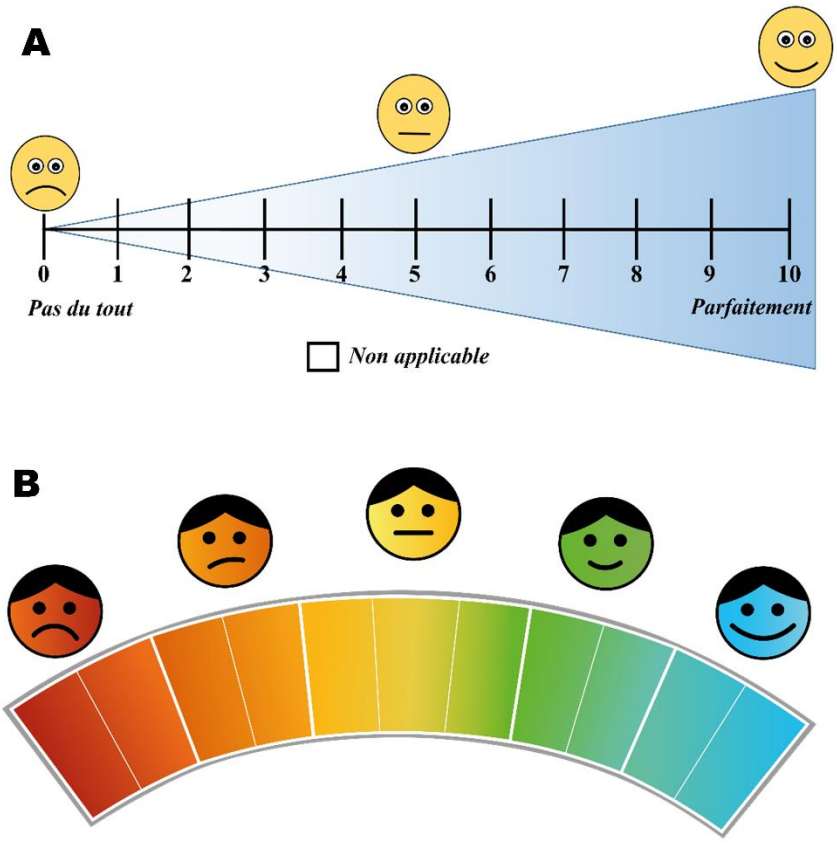
	Identify lateral movement (voice or footsteps)	x	x	
	Identify approach or recede (voice or footsteps)	x		x
	Identify if vehicle is approaching/receding	x		
	Internal or external sounds	x		
	Sounds closer than expected			
	Sounds further away than expected			
	Sounds in expected location		x	
C-Qualities of hearing	Separation of two sounds**	x		x
	Sounds appearing jumbled			
	Music and voice as separate objects	x		
	Identify different people by voice	x	x	x
	Distinguish familiar music	x	x	x
	Distinguish different sounds	x	x	x
	Identify instruments in music			
	Naturalness of music		x	
	Clarity of everyday sounds		x	
	Naturalness of other voices			
	Naturalness of everyday sounds			
	Naturalness of own voice			
	Judging mood from voice	x		
	Need to concentrate when listening	x		
	Effort of conversation	x		
	When you are a driver			
	When you are a passenger	x		
	Ability to ignore competing sounds	x		

*This item corresponds to:
-“locate someone familiar in the house” in the SSQ33
-“locate loud sounds in the house” in the Kid-SSQ
**This item corresponds to: “recognize different familiar sounds in the house” for the Kid-SSQ

Appendix 3: Number of items from each auditory situation category (from [23]*) included in the three versions of the SSQ (SSQ33. 15iSSQ. Kid-SSQ) and item numbers for the* Kid-SSQ shown in the right-most column.

Sub-scale	Categories	Number of items in SSQ33 pediatric long-form	Number of items in 15iSSQ adult short-form	Number of items in Kid-SSQ pediatric short-form	Corresponding item numbers in Kid-SSQ
A	Speech in quiet	2	0	0	
	Speech in noise	4	4	3	A.1 ; A.2 ; A.3
	Speech in speech contexts	2	1	1	A.4
	Multiple speech stream and switching	1	0	0	
B	Localization	5	3	2	B.1 ; B.2
	Distance and movement	7	2	2	B.3 ; B.4
C	Identification of sounds	4	3	4	C.1 ; C.2 ; C.3 ; C.4
	Segregation of sounds	2	0	0	
	Sound quality and naturalness	0	2	0	
	Listening effort	3	0	0	

Appendix 4: Kid-SSQ rating scale. (A) Numerical visual scale (pas du tout = I can't do it at all; parfaitement = I can do it perfectly); (B) Face scale - this scale is best used for 7 to 9 year-old children. Each box implicitly corresponds to a number (e.g. the first box to 0, the last to 10) allowing rapid scoring by the experimenter.



Appendix 5: Kid-SSQ, the original French version

Section A – Audition de la parole

1. Tu discutes avec quelqu'un, la TV est allumée dans la même pièce.

Peux-tu comprendre ce qu'il te dit sans baisser le son ?

2. Tu es assis autour d'une table avec un groupe de 5 personnes environ. L'endroit est bruyant, comme une salle de classe où les élèves se déplacent et discutent. Tu peux voir toutes les personnes autour de la table.

Peux-tu comprendre ce que les personnes du groupe disent ?

3. Tu es assis autour d'une table avec un groupe de 5 personnes environ. L'endroit est bruyant, comme une salle de classe où les élèves se déplacent et discutent. Tu ne peux pas voir toutes les personnes autour de la table.

Peux-tu comprendre ce que les personnes du groupe disent ?

4. Tu discutes avec quelqu'un dans une pièce où beaucoup d'autres personnes parlent.

Peux-tu comprendre ce qu'il te dit ?

Section B – Audition spatiale

1. Tu es à la maison et tu entends un bruit fort (une porte qui claque ou l'aspirateur).

Peux-tu dire immédiatement d'où vient le bruit ?

2. Tu es dehors. Un chien aboie très fort.

Peux-tu dire immédiatement où il se trouve, sans le voir ?

3. Tu es sur le trottoir d'une rue où il y a beaucoup de bruit. Tu entends un bus ou un camion.

Peux-tu dire immédiatement d'où il vient avant de le voir ?

4. Tu entends des voix ou des bruits de pas.

Juste en écoutant, peux-tu dire où la personne va ? (par exemple, est-ce que la personne se rapproche de toi ou se déplace de ta gauche vers ta droite ?)

<u>Subscale C – Qualités d'audition</u>

1. **Peux-tu reconnaître facilement la voix des personnes de ta famille, de tes amis, quand tu les entends arriver sans les voir ?**

2. Tu entends une chanson ou une musique que tu connais bien.
Arrives-tu facilement à la reconnaître ?

3. **Peux-tu faire la différence entre deux sons qui se ressemblent, comme par exemple un robinet qui coule et un ventilateur ?**

4. Tous les jours, tu entends les mêmes bruits à la maison.
Est-ce que tu peux les reconnaître facilement ? (par exemple, le bruit de la cafetière, la sonnerie du téléphone, l'eau qui coule…)

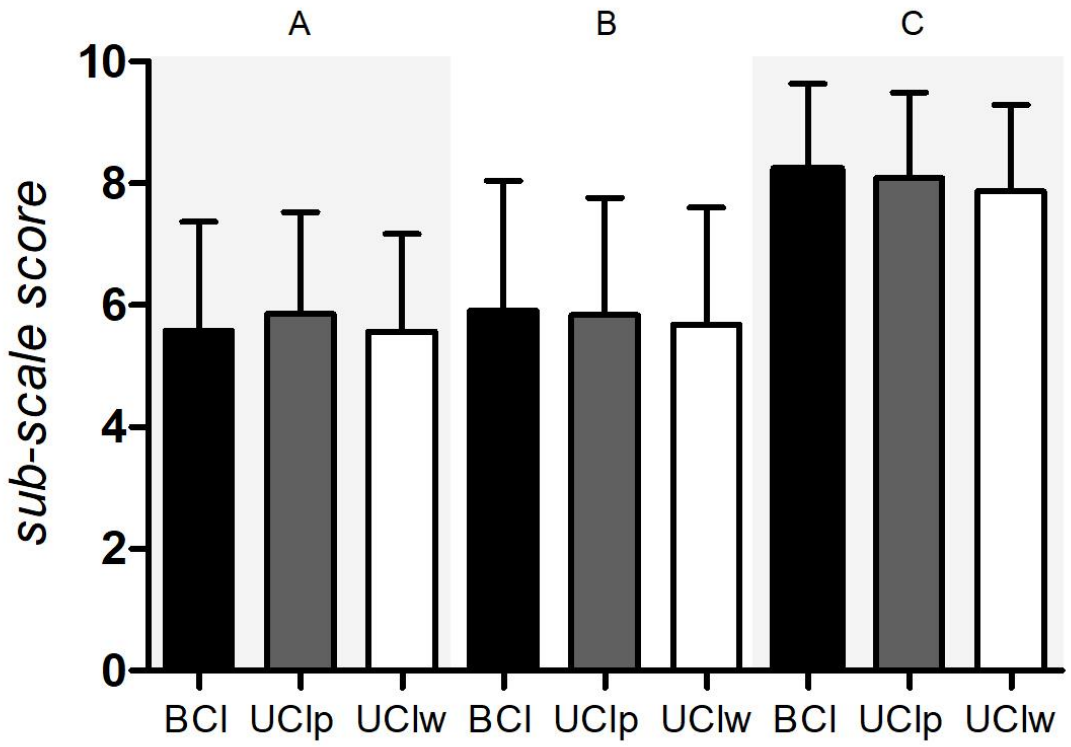
Appendix 6: Methodological details of the exploratory factor analysis of the Kid-SSQ.

Exploratory factor analysis was performed using a factor extraction performed on a non-parametric correlation matrix with a maximum likelihood method and an oblique rotation method (oblimin). The Kaiser-Meyer-Olkin measure of sampling adequacy was fair, with an overall result of 0.73 and all values for individual items were greater than 0.58. Bartlett's test was highly statistically significant (chi-squared=472, $p<10^{-60}$), confirming that correlations between individual items were sufficient. Cattell's scree test and the more objective parallel analysis gave 3 factors with an adjusted eigenvalue above 1. This 3 factors internal structure of the Kid-SSQ was then confirmed using Revelle's clustering method [39], which first identifies pairs of items with the greatest similarity, and continues by adding other items, one-by-one, with the aim of improving both internal consistency (measured by the alpha coefficient) and homogeneity (explored by Revelle's beta coefficient).

Appendix 7: Means (standard deviations) for each subscale and item shown separately for the BCI and UCI groups.

	A - Speech perception					B - Spatial hearing					C - Qualities of hearing				
	A.1	A.2	A.3	A.4	Total A	B.1	B.2	B.3	B.4	Total B	C.1	C.2	C.3	C.4	Total C
BCI n=100	6.8 (2.3)	5.5 (2.6)	3.5 (2.4)	6.5 (2.3)	**5.6 (1.8)**	6.5 (2.6)	6.3 (2.9)	5.6 (2.7)	5.2 (2.7)	**6.0 (2.1)**	8.7 (1.8)	8.4 (2.2)	7.3 (2.5)	8.5 (1.9)	**8.3 (1.4)**
UCI n=54	6.7 (2.1)	5.8 (2.2)	3.7 (2.4)	7.2 (2.4)	**5.9 (1.7)**	6.8 (3.0)	5.8 (2.8)	5.6 (2.7)	5.2 (2.8)	**5.8 (1.9)**	8.2 (2.2)	8.6 (1.7)	6.9 (2.9)	8.6 (1.5)	**8.1 (1.4)**

Appendix 8: Means and standard deviations on the three subscales for the Bilateral Cochlear Implant (BCI; black; n=100), Unilateral Cochlear Implant bimodal configuration (UCIp; grey; n =36) and Unilateral Cochlear Implant monaural configuration (UCIw; white; n = 18) children. (A: speech perception; B: spatial hearing; C: qualities of hearing).



BCI: bilateral cochlear implant; UCIp: unilateral cochlear implant with contralateral hearing prosthesis; UCIw: unilateral cochlear implant without contralateral hearing prosthesis.

Appendix 9: Results of consistency and internal structure analysis of the Kid-SSQ from a population of 154 children with cochlear implants.

On the left, results of factor analyses showing the factor loadings of each factor obtained after oblique rotation.

On the right, consistency measures and Cronbach's alpha values. The right-most column shows Cronbach's alpha for each subscale with the corresponding confidence interval underneath.

Kid-SSQ	**Standardized loadings (pattern matrix)**				**Consistency and Reliability**		
Items	**Factor 1**	**Factor 3**	**Factor 2**	**Communalities**	**Item to total correlation**	**Cronbach's Alpha without item**	**Cronbach's Alpha**
AQ1	**0.39**	0.25		0.26	0.42	0.76	0.73 (0.69; 0.76)
AQ2	**0.78**			0.58	0.41	0.77	
AQ3	**0.99**			0.99	0.53	0.75	
AQ4	0.21	0.39		0.25	0.44	0.76	
BQ1		0.11	**0.68**	0.53	0.51	0.75	0.71 (0.65; 0.75)
BQ2			**0.78**	0.57	0.45	0.76	
BQ3	0.14	0.29	**0.36**	0.36	0.52	0.75	
BQ4		0.28	**0.32**	0.23	0.36	0.77	
CQ1		**0.33**	0.28	0.26	0.40	0.77	0.56 (0.49; 0.61)
CQ2		**0.46**		0.21	0.22	0.78	
CQ3		**0.57**		0.31	0.36	0.77	
CQ4		**0.69**		0.48	0.43	0.77	