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QUALITY OF LIFE IN CHILDREN WITH HEARING IMPAIRMENT: SYSTEMATIC REVIEW AND META-ANALYSIS

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Abstract

Objective—To determine the impact of pediatric hearing loss on quality of life (QOL).

Data Sources—A qualified medical librarian conducted a literature search for relevant publications that evaluate QOL in school-aged children with hearing loss (HL).

Review Methods—Studies were assessed independently by two reviewers for inclusion in the systematic review and meta-analysis.

Results—From 979 abstracts, 69 were identified as relevant; ultimately 41 articles were included in the systematic review. This review revealed that children with HL generally report a lower QOL than their normal hearing peers, and QOL improves after interventions. The extent of these differences is variable among studies, and depends on the QOL measure. Four studies using the Pediatric Quality of Life Inventory (PedsQL) had sufficient data for inclusion in a meta-analysis. After pooling studies, statistically and clinically significant differences in PedsQL scores were found between children with normal hearing and those with HL, specifically in the Social and School domains. Statistically significant differences were also noted in in total scores for children with unilateral HL and in the physical domain for children with bilateral HL as compared to normal hearing, however these differences were not clinically meaningful.

Conclusions—Our analysis reveals that decreased QOL in children with HL is detected in distinct domains of the PedsQL questionnaire. These domains of school functioning and social interactions are especially important for development and learning. Future work should focus on these specific aspects of QOL when assessing HL in the pediatric population.

Keywords

quality of life; pediatric hearing loss

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Conflicts of interest: none

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INTRODUCTION

Hearing impairment is common among children and adolescents in the United States. A recent systematic review reported the incidence of neonatal hearing loss in the US to be 1.1 per 1000 infants and the average prevalence of mild or worse unilateral or bilateral hearing impairment in children and adolescents to be over 3%.[1] Hearing impairment puts children at a clear disadvantage. In addition to being 21–39% less likely to attend college, persons with hearing loss experience twice as much work stress and have lower labor participation rates than normal hearing individuals.[2] While it is clear that hearing loss is disabling in some ways, the true effect on quality of life remains unknown.

The World Health Organization defines quality of life (QOL) as *individuals' perception of their position in life in the context of culture and value systems in which they live and in relation to their goals, expectations, standards and concerns*.[3] The definition encompasses six domains of quality of life including a physical domain, psychological domain, level of independence, social relationships, environment, and spirituality/religion/personal beliefs. There are multiple QOL assessment tools that have been used to evaluate pediatric hearing impairment, however few were designed to specifically address this common problem. Additionally, no single tool is routinely used to report quality of life in children with hearing loss, resulting in variability and inconsistencies between studies reporting outcomes in these patients. While some prior studies have suggested decreased quality of life in pediatric patients with hearing loss, others have shown similar findings in children with hearing impairment as compared to those with normal hearing. The true effect of unilateral and bilateral hearing loss on QOL in children remains unclear.

The objective of this study was to assess the impact of pediatric hearing loss on quality of life by reviewing available studies on this topic. We hypothesized that quality of life was negatively affected in children diagnosed with hearing loss as compared to their normal hearing peers. A secondary objective was to evaluate the expected improvement in quality of life after intervention.

METHODS

The PRISMA guidelines were consulted and all requirements have been met for this study.[4] The published literature was searched using strategies created by a medical librarian for the concepts of "quality of life," "hearing loss" and "children." Abstracts that involved ongoing temporary or fluctuating conductive HL, such as that due to otitis media, were excluded, as were studies involving children with cognitive impairments. Only studies that used health-related or hearing-related quality of life instruments that had been previously validated in the literature, were included. These strategies were established using a combination of standardized terms and key words, and were implemented in PubMed 1946-, Embase 1947-, CINAHL, the Cochrane Database of Systematic Reviews, Cochrane Database of Abstracts of Review Effects (DARE), Cochrane Central Register of Controlled Trials (CENTRAL), clinicaltrials.gov, Proquest Dissertations and Theses, and FirstSearch Proceedings. Searches were limited to English using database supplied filters. The search was completed in June,

2014, exported to EndNote, and duplicates were removed. Our comprehensive search terms are available in the appendix.

Candidate articles were independently reviewed by two authors familiar with the subject material, and discrepancies were discussed amongst the two authors. Articles were considered eligible if they included pediatric patients with hearing loss who were assessed using a validated quality of life measure. Information was collected on a data extraction form created by the authors. Variables collected included quality of life measure used, number of patients included, age range of patients, and effect size of the outcome measure. Effect size was generally a comparison of means between two groups, or a point estimate of change in pre/post intervention studies.

Quality Assessment

Quality was assessed using a scale modified from the Newcastle-Ottawa Scale.[5] Three factors were rated as either low, high or unclear risk. The 3 factors included selection bias, outcome bias and overall risk of bias. Selection bias was determined based on representativeness of the population and demographics of the non-respondents as compared to the respondents. Outcome was assessed based on the use of a validated measurement tool, and the use of an appropriate statistical test. Studies were rated as “unclear” if details of these categories were not specified in the manuscript. Due to small sample size, studies were not excluded from the meta-analyses due to high or unclear risk of bias.

Meta-analysis

A meta-analysis was performed for quality of life measures with more than 2 studies reporting outcomes. Studies were required to contain both children with hearing loss and those with normal hearing (control group), and were separated based on unilateral or bilateral hearing loss. Further analyses were done to evaluate distinct quality of life domains. For our secondary outcome, a meta-analysis was performed for measures with more than 2 studies comparing quality of life pre- and post-treatment. A random effects analysis was performed for all analyses. Analyses combining data from multiple studies were performed using STATA statistical software version 13.1 (STATA Corporation, College Station, TX).

Clinical significance was determined based on previous studies of these validated tools. Clinical significance for the PedsQL tool has not been previously investigated in children with hearing loss. However this tool has been previously interrogated in a very large study of over 4,000 pediatric patients with diabetes. The minimal clinically important difference (MCID) scores for this quality of life measure was found to be dependent on type of diabetes, as well as parent versus child report, and ranged between 4 and 6 points on the PedsQL survey.[6] This reference range was used when analyzing the results from our current study, and results were considered clinically significant if the absolute value of the effect size was 4 points or greater.

RESULTS

Description of studies

Following our comprehensive search and exclusion of duplicate studies, 1068 articles were identified. After reviewing titles, 979 abstracts were assessed for complete review of text. Sixty-nine complete articles were read, and 37 studies met criteria for inclusion. An additional 5 articles were identified during the review process and were included in the study. One article was removed due to use of duplicate data and patients. Ultimately, we included 41 articles in our systematic review (Figure 1). Information for the included studies is shown in tables 2–4.

Outcome measures

There were multiple assessment tools used to assess quality of life in these pediatrics patients, ranging from generic quality of life measurements to those specifically assessing aspects of hearing loss (tables 2–4). The most commonly used assessment tools in this review are described below.

Glasgow Children's Benefit Inventory (GCBI)[7]—This tool is used to assess benefit retrospectively after an intervention. There are 4 domains: emotion, physical health, learning and vitality. Answers are provided on a 5 point likert scale and converted to a scale ranging from −100 to +100. Higher positive scores indicate more improvement.

Health Utilities Index Mark 3 (HUI3)[8,9]—This tool is a utility-based measure of overall quality of life. There are 8 domains including vision, hearing, speech, ambulation, dexterity, emotion, cognition and pain. Higher scores indicate higher quality of life.

Pediatric Quality of Life Inventory (PedsQL)[10]—This measurement of overall health related quality of life is rated on a 5-point likert scale and converted to a scale from 0–100. There are 4 domains including physical, emotion, social and school. Higher scores indicate higher quality of life.

Hearing Environments and Reflection on Quality of Life Questionnaire (HEAR-QL)[11]—This tool is comprised of 26 questions focused on 3 domains, situations affecting interaction with family and friends, participation in school and social activities and the impact of impaired hearing on emotional well-being. Higher scores indicate higher quality of life.

Speech, Spatial and Qualities of Hearing Scale (SSQ)[12]—This tool has 3 domains, speech, spatial and qualities of hearing. The measurement was designed to evaluate the effects of hearing impairment specifically, with higher scores representing less disability.

Quality assessment

Quality assessment is shown in Table 1. Overall, there was low overall risk of bias in these studies. All studies used an appropriate statistical test and a validated quality of life metric although one study received an "unclear" rating for outcome bias as the quality of life metric

used was validated, but then slightly modified for use. Fifteen studies were rated “unclear” for the selection category due to lack of detail regarding the selection process or the demographics of the non-responding population. No studies were considered high risk for the selection category as they used appropriate methods for recruitment.

Systematic Review

Sixteen studies attempted to address the potential difference in quality of life between children with hearing impairment and their normal hearing peers (table 2). Ten of these studies directly surveyed normal hearing children as a comparison group and 6 used databases of normative scores for commonly used assessment tools. Of these 16 studies, 11 (6 using a control group and 5 using normative scores for comparison) identified statistical differences between patients with hearing loss and those with normal hearing using one or more assessment tools, or by specifically looking at individual domains of a questionnaire. The remaining 5 studies (4 using control group and 1 using normative scores) did not identify any differences in quality of life between the two pediatric groups.

There were 21 studies evaluating quality of life improvement following an intervention, such as CI, BAHA or hearing aid use (table 3). Ten studies evaluated children following BAHA or soft band, 2 studies assessed children after hearing aid fitting, 7 studies included patients following cochlear implantation and 2 studies addressed the benefit of a second cochlear implantation. Overall, using a variety of assessment tools, both general and those specific to hearing impairment, interventions were convincingly helpful in improving quality of life from both patient and parental perspectives.

Lastly, there were 7 additional studies addressing quality of life in pediatric patients with hearing loss (table 4). These studies varied in their objective from evaluating the timing of identification of hearing loss on quality of life to noting differences in self-reporting of quality of life based on age.

Meta analyses

Four studies using child-reported PedsQL scores had sufficient data for inclusion in a meta-analysis. After pooling studies, statistically and clinically significant differences in PedsQL scores were found between children with normal hearing and those with hearing loss, both unilateral and bilateral, specifically in the School domain (difference of 8.79 points, 95% CI: 4.03–13.55 for unilateral hearing loss and difference of 6.93 points, 95% CI: 3.47–10.40 for bilateral hearing loss) and the Social domain (difference of 4.31 points, 95% CI: 0.26–9.22 for unilateral hearing loss and difference of 4.31 points, 95% CI: 0.07–8.54 for bilateral hearing loss). Additionally, statistically significant differences were seen in total scores for children with unilateral hearing loss only (difference of 3.8 points, 95% CI: 0.2–7.4) and in the physical domain between normal hearing and those with bilateral hearing loss (difference of 3.15 points, 95% CI: 0.23–6.07) however these differences were not clinically significant (table 5, and figures 2–5). Clinically significant differences were considered those with an absolute value of 4 or more, as previous studies have identified an MCID of 4–6 points for the PedsQL.

The GCBI was the only quality of life measure with enough data for a combined analysis for pre/post intervention studies. A meta-analysis was used to combine the change in scores on GCBI following BAHA placement in children with both unilateral and bilateral hearing loss. Three studies included children with bilateral hearing loss only,[13–15] 1 study included children with unilateral hearing loss only,[16] and 1 study included children with both unilateral and bilateral hearing loss,[17] but with the data separated in the results. There were two other studies which used the GCBI in patients with BAHA placement,[18,19] however after reaching out to authors by email, the appropriate necessary data for inclusion in the meta-analysis was only available from a total of 5 studies. As all studies showed a positive change in GCBI scores, it was not surprising that the meta-analysis revealed a positive change when combining all children (bilateral and unilateral hearing loss) as well as when studying only children with bilateral hearing loss. The mean change in GCBI scores were +40.35 (95% CI 24.60–56.09) for all children with hearing loss, and +43.02 points (95% CI 24.91–61.13) for children with bilateral hearing loss only (figures 5–6). Unfortunately, clinical significance has not yet been determined for this quality of life measure, so while GCBI scores show statistically significant improvement, we are unable to comment on clinical significance.

DISCUSSION

In this study, we have shown that there is high variability in reported quality of life in children with hearing loss. Using the only measure with adequate studies for meta-analysis, we found that the only domains with both statistically and clinically significantly differences between children with hearing loss and those with normal hearing are school and social categories. In this analysis, the differences in physical and emotional domains were not found to be clinically significant. While these findings are intuitive, our study is the first to show this in a comprehensive fashion.

A second aim of this study was to evaluate the potential improvement of quality of life in children following intervention. As seen in table 2, our review of the literature suggests that quality of life is significantly improved following aided hearing, BAHA and CI implantation. Two studies evaluated the effect of a second implant, however the results were inconsistent. In the larger study of the two, differences in quality of life were found only in disease-specific quality of life assessments, suggesting the importance of using measures which are specific to the hearing loss. Our meta-analysis combing GCBI scores in children after BAHA placement confirms that there is improvement in this measure following intervention, however we are unable to comment on clinical significance at this time, as it has not been determined for this quality of life tool.

There are multiple limitations of this study. The inherent limitations of a systematic review and meta-analysis exist, including that our analysis is dependent on the studies that our search returns. While statistical significance was addressed in several studies, few studies addressed the issue of clinical significance. As shown in our quality assessment, the risk of selection bias was often unclear, as many studies did not address the non-responders in the study, an important factor to consider in a survey study. A second limitation is that we are only able to perform a meta-analysis on quality of life measures with an adequate number of

studies, and the number of studies included is relatively low. Additionally, while the PedsQL tool has been validated for use for overall total scores and the physical domain, the emotional, social and school domains have not been validated separately, which limits the strength of the conclusions drawn from these results. A third and major weakness of this study is the variability in age, hearing level and type of hearing loss of the participants in these studies. Unfortunately, due to small sample size, we are unable to stratify based on level of hearing loss or age of patients. Lastly, the wide range of quality of life measures used in these studies makes them difficult to combine in an organized fashion. Multiple measures are used commonly in assessing quality of life in children suffering from various disabilities; however there is no single, accepted quality of life tool used for pediatric hearing loss. The need for such a tool is obvious as pediatric hearing loss is a common problem and there appears to be substantial effects on quality of life, at least in certain specific aspects of functioning.

While previous studies have evaluated the effect of hearing loss on quality of life in pediatric patients,[20,21] we believe that this is the first attempt to analytically differentiate between specific domains of quality of life measures. Previous work has been inconsistent in identifying quality of life deficits in children with hearing loss. One possible explanation for this is that while overall quality of life may not be clinically or statistically significantly affected in pediatric patients with hearing loss, there may be certain aspects of life in which quality is severely affected. The quality of life measures currently used may not be sensitive to these specific aspects. An instrument addressing the relevant domains of quality of life in hearing impaired children, such as the Youth Quality of Life Instrument (YQOL) and the Hearing Environments and Reflection on Quality of Life Questionnaire (HEAR-QL) may be more sensitive in detecting differences in hearing related quality of life. Thus far, the HEAR-QL has been shown to be valid and reliable as well as more sensitive to quality of life differences in children with hearing loss than the PedsQL, the most commonly used measure in our review.[11] HEAR-QL has only a moderate correlation to the PedsQL[22,23] and likely the addition of the “physical” domain in the PedsQL, as well as many other commonly used quality of life questionnaires, dilutes the clinical difference in quality of life when comparing children with hearing loss to their normal hearing peers. We believe that a focus on social interactions and school activities is especially important when considering quality of life in hearing impaired children as communication and learning are adversely affected in this patient population.

CONCLUSION

While there is variability in reported influences on quality of life from pediatric hearing loss, our analysis reveals that decreased quality of life in children with hearing loss is detected in distinct domains of the PedsQL questionnaire, school activities and social interactions. There is a need for a quality of life tool with a focus on these specific aspects when assessing hearing loss in the pediatric population.

Acknowledgments

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Appendix: Search Terms

Deaf

"Deafness"[Mesh] OR "Deafness" OR "Deaf" OR "Bilateral Deafness" OR "Complete Hearing Loss" OR "Prelingual Deafness" OR "Deaf-Mutism" OR "Deaf Mutism" OR "Acquired Deafness" OR "Extreme Hearing Loss"

Embase

'Deafness' OR 'Deaf' OR 'Bilateral Deafness' OR 'Complete Hearing Loss' OR 'Prelingual Deafness' OR 'Deaf-Mutism' OR 'Deaf Mutism' OR 'Acquired Deafness' OR 'Extreme Hearing Loss'

CINAHL

MH "Deafness+" OR "Deafness" OR "Deaf" OR "Bilateral Deafness" OR "Complete Hearing Loss" OR "Prelingual Deafness" OR "Deaf-Mutism" OR "Deaf Mutism" OR "Acquired Deafness" OR "Extreme Hearing Loss"

AND

Children, 5–18

PubMed

"Child"[Mesh] OR "Disabled Children"[Mesh] OR "Adolescent"[Mesh] OR "Child, Preschool"[Mesh] OR "Preschool Child" OR "Preschool Children" OR "Child" OR "Children" OR "Disabled Child" OR "Handicapped Children" OR "Children with Disability" OR "Children with Disabilities" OR "Adolescent" OR "Adolescents" OR "Teens" OR "Teen" OR "Teenagers" OR "Teenager" OR "Youth" OR "Youths" OR "Adolescence" OR "girl" OR "girls" OR "boy" OR "boys" OR "juvenile" OR "juveniles"

Embase

'pediatrics'/exp OR 'child'/exp OR 'adolescent'/exp OR 'Child' OR 'Children' OR 'Children' OR 'Disabled Child' OR 'Handicapped Children' OR 'Children with Disability' OR 'Children with Disabilities' OR 'Preschool Child' OR 'Preschool Children' OR 'Adolescent' OR 'Adolescents' OR Teen* OR 'Youth' OR 'Youths' OR 'Adolescence' OR 'girl' OR 'girls' OR 'boy' OR 'boys' OR 'juvenile' OR 'juveniles' OR 'Pediatrics' OR 'pediatric' OR 'pediatry' OR 'section 7'

CINAHL

MH "Child+" OR MH "Adolescence+" OR MH "Minors (Legal)" OR "Preschool Child" OR "Preschool Children" OR "Child" OR "Children" OR "Disabled Child" OR "Handicapped Children" OR "Children with Disability" OR "Children with Disabilities" OR

"Adolescent" OR "Adolescents" OR "Teens" OR "Teen" OR "Teenagers" OR "Teenager" OR "Youth" OR "Youths" OR "Adolescence" OR "girl" OR "girls" OR "boy" OR "boys" OR "juvenile" OR "juveniles"

AND

Quality of Life

PubMed

"Quality of Life"[Mesh] OR "Quality of Life" OR "Life Qualities" OR "Life Quality" OR "HRQL" OR "health related quality of life" OR "HRQOL"

Embase

'quality of life'/exp OR 'Quality of Life' OR 'Life Qualities' OR 'Life Quality' OR 'HRQL' OR 'health related quality of life' OR 'HRQOL'

CINAHL

MH "Quality of Life" OR "Quality of Life" OR "Life Qualities" OR "Life Quality" OR "HRQL" OR "health related quality of life" OR "HRQOL"

AND

Validated HRQL Instruments (Specific)

PubMed

(instrumentation[sh] OR Validation Studies[pt] OR "Reproducibility of Results"[Mesh] OR reproducib*[tiab] OR "Psychometrics"[Mesh] OR psychometr*[tiab] OR clinimetr*[tiab] OR clinometr*[tiab] OR "Observer Variation"[Mesh] OR "observer variation"[tiab] OR "Discriminant Analysis"[Mesh] OR reliab*[tiab] OR valid*[tiab] OR coefficient[tiab] OR "internal consistency"[tiab] OR "item correlation"[tiab] OR "item correlations"[tiab] OR "item selection"[tiab] OR "item selections"[tiab] OR "item reduction"[tiab] OR "item reductions"[tiab] OR agreement[tw] OR precision[tw] OR imprecision[tw] OR "precise values"[tw] OR test–retest[tiab] OR stability[tiab] OR interrater[tiab] OR inter-rater[tiab] OR intrarater[tiab] OR intra-rater[tiab] OR intertester[tiab] OR inter-tester[tiab] OR intratester[tiab] OR intra-tester[tiab] OR interobserver[tiab] OR inter-observer[tiab] OR intraobserver[tiab] OR intra-observer[tiab] OR intertechnician[tiab] OR intertechnician[tiab] OR intratechnician[tiab] OR intra-technician[tiab] OR interexaminer[tiab] OR inter-examiner[tiab] OR intraexaminer[tiab] OR intra-examiner[tiab] OR interassay[tiab] OR inter-assay[tiab] OR intraassay[tiab] OR intra-assay[tiab] OR interindividual[tiab] OR inter-individual[tiab] OR intraindividual[tiab] OR intra-individual[tiab] OR interparticipant[tiab] OR inter-participant[tiab] OR intraparticipant[tiab] OR intra-participant[tiab] OR kappa[tiab] OR kappa's[tiab] OR kappas[tiab] OR "coefficient of variation"[tiab] OR repeatab*[tw] OR generaliza*[tiab] OR generalisa*[tiab] OR concordance[tiab] OR discriminative[tiab] OR "known group"[tiab] OR "factor analysis"[tiab] OR "factor analyses"[tiab] OR "factor structure"[tiab] OR "factor structures"[tiab] OR

dimensionality[tiab] OR subscale*[tiab] OR “multitrait scaling analysis”[tiab] OR “multitrait scaling analyses”[tiab] OR “item discriminant”[tiab]OR “interscale correlation” [tiab] OR “interscale correlations”[tiab] OR “individual variability”[tiab] OR “interval variability”[tiab] OR “rate variability”[tiab] OR “variability analysis”[tiab] OR “standard error of measurement”[tiab] OR sensitiv*[tiab] OR responsive*[tiab] OR “minimal detectable concentration”[tiab] OR interpretab*[tiab] OR “meaningful change”[tiab] OR “minimal important change”[tiab] OR “minimal important difference”[tiab] OR “minimally important change”[tiab] OR “minimally important difference”[tiab] OR “minimal detectable change”[tiab] OR “minimal detectable difference”[tiab] OR “minimally detectable change” [tiab] OR “minimally detectable difference”[tiab] OR “minimal real change”[tiab] OR “minimal real difference”[tiab] OR “minimally real change”[tiab] OR “minimally real difference”[tiab] OR “ceiling effect”[tiab] OR “floor effect”[tiab] OR “Item response model”[tiab] OR IRT[tiab] OR Rasch[tiab] OR “Differential item functioning”[tiab] OR DIF[tiab] OR “computer adaptive testing”[tiab] OR “item bank”[tiab] OR “cross-cultural equivalence”[tiab])

Embase

‘instrumentation’/exp OR ‘validation study’/exp OR ‘reproducibility’/exp OR reproducib* OR ‘psychometry’/exp OR psychometr*OR clinimetr*OR clinometr* OR ‘observer variation’/exp OR ‘observer variation’ OR ‘discriminant analysis’/exp OR reliab* OR valid* OR coefficient OR ‘internal consistency’ OR ‘item correlation’ OR ‘item correlations’ OR ‘item selection’ OR ‘item selections’ OR ‘item reduction’ OR ‘item reductions’ OR agreement OR precision OR imprecision OR ‘precise values’ OR test–retest OR stability OR interrater OR inter-rater OR intrarater OR intra-rater OR intertester OR inter-tester OR intratester OR intra-tester OR interobserver OR inter-observer OR intraobserver OR intra-observer OR intertechnician OR intertechnician OR intratechnician OR intra-technician OR interexaminer OR inter-examiner OR intraexaminer OR intra-examiner OR interassay OR inter-assay OR intraassay OR intra-assay OR interindividual OR inter-individual OR intraindividual OR intra-individual OR interparticipant OR inter-participant OR intraparticipant OR intra-participant OR kappa OR kappas OR ‘coefficient of variation’ OR repeatab* OR generaliza* OR generalisa* OR concordance OR discriminative OR ‘known group’ OR ‘factor analysis’ OR ‘factor analyses’ OR ‘factor structure’ OR ‘factor structures’ OR dimensionality OR subscale* OR ‘multitrait scaling analysis’ OR ‘multitrait scaling analyses’ OR ‘item discriminant’ OR ‘interscale correlation’ OR ‘interscale correlations’ OR ‘individual variability’ OR ‘interval variability’ OR ‘rate variability’ OR ‘variability analysis’ OR ‘standard error of measurement’ OR sensitiv* OR responsive* OR ‘minimal detectable concentration’ OR interpreta* OR ‘meaningful change’ OR ‘minimal important change’ OR ‘minimal important difference’ OR ‘minimally important change’ OR ‘minimally important difference’ OR ‘minimal detectable change’ OR ‘minimal detectable difference’ OR ‘minimally detectable change’ OR ‘minimally detectable difference’ OR ‘minimal real change’ OR ‘minimal real difference’ OR ‘minimally real change’ OR ‘minimally real difference’ OR ‘ceiling effect’ OR ‘floor effect’ OR ‘Item response model’ OR IRT OR Rasch OR ‘Differential item functioning’ OR DIF OR ‘computer adaptive testing’ OR ‘item bank’ OR ‘cross cultural equivalence’

CINAHL

MH “Validation Studies” OR MH “Reproducibility of Results” OR MH “Psychometrics” OR psychometr*OR clinimetr*OR clinometr* OR “observer variation”/exp OR “observer variation” OR “discriminant analysis” OR reliab* OR valid* OR coefficient OR “internal consistency” OR “item correlation” OR “item correlations” OR “item selection” OR “item selections” OR “item reduction” OR “item reductions” OR agreement OR precision OR imprecision OR “precise values” OR test–retest OR stability OR interrater OR inter-rater OR intrarater OR intra-rater OR intertester OR inter-tester OR intratester OR intra-tester OR interobserver OR inter-observer OR intraobserver OR intra-observer OR intertechnician OR intertechnician OR intratechnician OR intra-technician OR interexaminer OR inter-examiner OR intraexaminer OR intra-examiner OR interassay OR inter-assay OR intraassay OR intra-assay OR interindividual OR inter-individual OR intraindividual OR intra-individual OR interparticipant OR inter-participant OR intraparticipant OR intra-participant OR kappa OR kappas OR “coefficient of variation” OR repeatab* OR generaliza* OR generalisa* OR concordance OR discriminative OR “known group” OR “factor analysis” OR “factor analyses” OR “factor structure” OR “factor structures” OR dimensionality OR subscale* OR “multitrait scaling analysis” OR “multitrait scaling analyses” OR “item discriminant” OR “interscale correlation” OR “interscale correlations” OR “individual variability” OR “interval variability” OR “rate variability” OR “variability analysis” OR “standard error of measurement” OR sensitiv* OR responsive* OR “minimal detectable concentration” OR interpreta* OR “meaningful change” OR “minimal important change” OR “minimal important difference” OR “minimally important change” OR “minimally important difference” OR “minimal detectable change” OR “minimal detectable difference” OR “minimally detectable change” OR “minimally detectable difference” OR “minimal real change” OR “minimal real difference” OR “minimally real change” OR “minimally real difference” OR “ceiling effect” OR “floor effect” OR “Item response model” OR IRT OR Rasch OR “Differential item functioning” OR DIF OR “computer adaptive testing” OR “item bank” OR “cross cultural equivalence”

NOT

Cognitive impairment

PubMed—“cognition disorder” OR “cognition disorders” OR “cognitive defects” OR “cognitive deficit” OR “cognitive disability” OR “cognitive disorder” OR “cognitive disorders” OR “cognitive dysfunction” OR “cognitive impairment” OR “delirium” OR “dementia” OR “amnestic” OR “cognitive disorders” OR “overinclusion” OR “response interference” OR “Cognition Disorders”[Mesh] OR “Cognition Disorders” OR “Auditory Perceptual Disorder” OR “Psychoacoustical Disorders” OR “Psychoacoustical Disorder” OR “Auditory Comprehension Disorder” OR “Auditory Comprehension Disorders” OR “Auditory Processing Disorder” OR “Auditory Processing Disorders” OR “Acoustic Perceptual Disorder” OR “Acoustic Perceptual Disorders” OR “Auditory Inattention” OR “Auditory Inattentions” OR “Huntington Disease” OR “Huntington Chorea” OR “Huntington’s Disease” OR “Huntington Chronic Progressive Hereditary Chorea” OR “Chronic Progressive Hereditary Chorea” OR “Huntington’s Chorea” OR “Late-Onset Huntington Disease” OR “Late Onset Huntington Disease” OR “Juvenile Huntington

Disease" OR "Juvenile-Onset Huntington Disease" OR "Juvenile Onset Huntington Disease" OR "Akinetic-Rigid Variant of Huntington Disease" OR "Akinetic Rigid Variant of Huntington Disease" OR "Dementia"OR "Delirium" OR "Amnesia" OR "Dementias" OR "Amentia" OR "Amentias" OR "Deliriums"

Embase—'cognition disorder' OR 'cognition disorders' OR 'cognitive defects' OR 'cognitive deficit' OR 'cognitive disability' OR 'cognitive disorder' OR 'cognitive disorders' OR 'cognitive dysfunction' OR 'cognitive impairment' OR 'delirium' OR 'dementia' OR 'amnestic' OR 'cognitive disorders' OR 'overinclusion' OR 'response interference' OR 'Cognition Disorders' OR 'Cognition Disorders' OR 'Auditory Perceptual Disorder' OR 'Psychoacoustical Disorders' OR 'Psychoacoustical Disorder' OR 'Auditory Comprehension Disorder' OR 'Auditory Comprehension Disorders' OR 'Auditory Processing Disorder' OR 'Auditory Processing Disorders' OR 'Acoustic Perceptual Disorder' OR 'Acoustic Perceptual Disorders' OR 'Auditory Inattention' OR 'Auditory Inattentions' OR 'Huntington Disease' OR 'Huntington Chorea' OR 'Huntington's Disease' OR 'Huntington Chronic Progressive Hereditary Chorea' OR 'Chronic Progressive Hereditary Chorea' OR 'Huntington's Chorea' OR 'Late-Onset Huntington Disease' OR 'Late Onset Huntington Disease' OR 'Juvenile Huntington Disease' OR 'Juvenile-Onset Huntington Disease' OR 'Juvenile Onset Huntington Disease' OR 'Akinetic-Rigid Variant of Huntington Disease' OR 'Akinetic Rigid Variant of Huntington Disease' OR 'Dementia'OR 'Delirium' OR 'Amnesia' OR 'Dementias' OR 'Amentia' OR 'Amentias' OR 'Deliriums'

CINAHL—"cognition disorder" OR "cognition disorders" OR "cognitive defects" OR "cognitive deficit" OR "cognitive disability" OR "cognitive disorder" OR "cognitive disorders" OR "cognitive dysfunction" OR "cognitive impairment" OR "delirium" OR "dementia" OR "amnestic" OR "cognitive disorders" OR "overinclusion" OR "response interference" OR "Cognition Disorders" OR "Cognition Disorders" OR "Auditory Perceptual Disorder" OR "Psychoacoustical Disorders" OR "Psychoacoustical Disorder" OR "Auditory Comprehension Disorder" OR "Auditory Comprehension Disorders" OR "Auditory Processing Disorder" OR "Auditory Processing Disorders" OR "Acoustic Perceptual Disorder" OR "Acoustic Perceptual Disorders" OR "Auditory Inattention" OR "Auditory Inattentions" OR "Huntington Disease" OR "Huntington Chorea" OR "Huntington"s Disease" OR "Huntington Chronic Progressive Hereditary Chorea" OR "Chronic Progressive Hereditary Chorea" OR "Huntington"s Chorea" OR "Late-Onset Huntington Disease" OR "Late Onset Huntington Disease" OR "Juvenile Huntington Disease" OR "Juvenile-Onset Huntington Disease" OR "Juvenile Onset Huntington Disease" OR "Akinetic-Rigid Variant of Huntington Disease" OR "Akinetic Rigid Variant of Huntington Disease" OR "Dementia"OR "Delirium" OR "Amnesia" OR "Dementias" OR "Amentia" OR "Amentias" OR "Deliriums"

NOT

PubMed

"Tinnitus"[Mesh] OR "Otitis Media"[Mesh] OR "Tinnitus" OR "Middle Ear Inflammation" OR "ear buzzing" OR "false latent otitis" OR "middle ear infection" OR "middle ear infections" OR "tympanitis"

Embase

'tinnitus'/exp OR 'otitis media'/exp OR 'tinnitus' OR 'Middle Ear Inflammation' OR 'ear buzzing' OR 'false latent otitis' OR 'middle ear infection' OR 'middle ear infections' OR 'tympanitis'

CINAHL

MH "Tinnitus" OR MH "Otitis Media" OR "Tinnitus" OR "Middle Ear Inflammation" OR "ear buzzing" OR "false latent otitis" OR "middle ear infection" OR "middle ear infections" OR "tympanitis"

NOT

("addresses"[Publication Type] OR "biography"[Publication Type] OR "case reports" [Publication Type] OR "comment"[Publication Type] OR "directory"[Publication Type] OR "editorial"[Publication Type] OR "festschrift"[Publication Type] OR "interview" [Publication Type] OR "lectures"[Publication Type] OR "legal cases"[Publication Type] OR "legislation"[Publication Type] OR "letter"[Publication Type] OR "news"[Publication Type] OR "newspaper article"[Publication Type] OR "patient education handout"[Publication Type] OR "popular works"[Publication Type] OR "congresses"[Publication Type] OR "consensus development conference"[Publication Type] OR "consensus development conference, nih"[Publication Type] OR "practice guideline"[Publication Type])

NOT

Animals

PubMed—("animals"[Mesh] NOT "humans"[Mesh])

Embase—([animals]/lim NOT [humans]/lim)

C&P PubMed (22)

("Deafness"[Mesh] OR "Deafness" OR "Deaf" OR "Bilateral Deafness" OR "Complete Hearing Loss" OR "Prelingual Deafness" OR "Deaf-Mutism" OR "Deaf Mutism" OR "Acquired Deafness" OR "Extreme Hearing Loss") AND ("Child" [Mesh] OR "Infant" [Mesh] OR "Adolescent" [Mesh] OR "Pediatrics" [Mesh] OR Child OR children OR Adolescen* OR Teen* OR Youth OR Youths OR girl* OR boy OR boys OR juvenile* OR pediatrics OR pediatric OR pediatry OR "section 7" OR "disabled child" OR "handicapped child" OR "children with disability" OR "children with disabilities") AND ("Quality of Life"[Mesh] OR "Quality of Life" OR "Life Qualities" OR "Life Quality" OR "HRQL" OR

"health related quality of life" OR "HRQOL") AND (instrumentation[sh] OR Validation Studies[pt] OR "Reproducibility of Results"[Mesh] OR reproducib*[tiab] OR "Psychometrics"[Mesh] OR psychometr*[tiab] OR clinimetr*[tiab] OR clinometr*[tiab] OR "Observer Variation"[Mesh] OR "observer variation"[tiab] OR "Discriminant Analysis" [Mesh] OR reliab*[tiab] OR valid*[tiab] OR coefficient[tiab] OR "internal consistency" [tiab] OR "item correlation"[tiab] OR "item correlations"[tiab] OR "item selection"[tiab] OR "item selections"[tiab] OR "item reduction"[tiab] OR "item reductions"[tiab] OR agreement[tw] OR precision[tw] OR imprecision[tw] OR "precise values"[tw] OR test–retest[tiab] OR stability[tiab] OR interrater[tiab] OR inter-rater[tiab] OR intrarater[tiab] OR intra-rater[tiab] OR intertester[tiab] OR inter-tester[tiab] OR intratester[tiab] OR intra-tester[tiab] OR interobserver[tiab] OR inter-observer[tiab] OR intraobserver[tiab] OR intra-observer[tiab] OR intertechnician[tiab] OR intertechnician[tiab] OR intratechnician[tiab] OR intra-technician[tiab] OR interexaminer[tiab] OR inter-examiner[tiab] OR intraexaminer[tiab] OR intra-examiner[tiab] OR interassay[tiab] OR inter-assay[tiab] OR intraassay[tiab] OR intra-assay[tiab] OR interindividual[tiab] OR inter-individual[tiab] OR intraindividual[tiab] OR intra-individual[tiab] OR interparticipant[tiab] OR inter-participant[tiab] OR intraparticipant[tiab] OR intra-participant[tiab] OR kappa[tiab] OR kappa's[tiab] OR kappas[tiab] OR "coefficient of variation"[tiab] OR repeatab*[tw] OR generaliza*[tiab] OR generalisa*[tiab] OR concordance[tiab] OR discriminative[tiab] OR "known group"[tiab] OR "factor analysis"[tiab] OR "factor analyses"[tiab] OR "factor structure"[tiab] OR "factor structures"[tiab] OR dimensionality[tiab] OR subscale*[tiab] OR "multitrait scaling analysis"[tiab] OR "multitrait scaling analyses"[tiab] OR "item discriminant"[tiab]OR "interscale correlation"[tiab] OR "interscale correlations"[tiab] OR "individual variability"[tiab] OR "interval variability"[tiab] OR "rate variability"[tiab] OR "variability analysis"[tiab] OR "standard error of measurement"[tiab] OR sensitiv*[tiab] OR responsive*[tiab] OR "minimal detectable concentration"[tiab] OR interpretab*[tiab] OR "meaningful change"[tiab] OR "minimal important change"[tiab] OR "minimal important difference"[tiab] OR "minimally important change"[tiab] OR "minimally important difference"[tiab] OR "minimal detectable change"[tiab] OR "minimal detectable difference" [tiab] OR "minimally detectable change"[tiab] OR "minimally detectable difference"[tiab] OR "minimal real change"[tiab] OR "minimal real difference"[tiab] OR "minimally real change"[tiab] OR "minimally real difference"[tiab] OR "ceiling effect"[tiab] OR "floor effect"[tiab] OR "Item response model"[tiab] OR IRT[tiab] OR Rasch[tiab] OR "Differential item functioning"[tiab] OR DIF[tiab] OR "computer adaptive testing"[tiab] OR "item bank"[tiab] OR "cross-cultural equivalence"[tiab]) NOT ("cognition disorder" OR "cognition disorders" OR "cognitive defects" OR "cognitive deficit" OR "cognitive disability" OR "cognitive disorder" OR "cognitive disorders" OR "cognitive dysfunction" OR "cognitive impairment" OR "delirium" OR "dementia" OR "amnestic" OR "cognitive disorders" OR "overinclusion" OR "response interference" OR "Cognition Disorders" [Mesh] OR "Cognition Disorders" OR "Auditory Perceptual Disorder" OR "Psychoacoustical Disorders" OR "Psychoacoustical Disorder" OR "Auditory Comprehension Disorder" OR "Auditory Comprehension Disorders" OR "Auditory Processing Disorder" OR "Auditory Processing Disorders" OR "Acoustic Perceptual Disorder" OR "Acoustic Perceptual Disorders" OR "Auditory Inattention" OR "Auditory Inattentions" OR "Huntington Disease" OR "Huntington Chorea" OR "Huntington's

Disease” OR “Huntington Chronic Progressive Hereditary Chorea” OR “Chronic Progressive Hereditary Chorea” OR “Huntington’s Chorea” OR “Late-Onset Huntington Disease” OR “Late Onset Huntington Disease” OR “Juvenile Huntington Disease” OR “Juvenile-Onset Huntington Disease” OR “Juvenile Onset Huntington Disease” OR “Akinetic-Rigid Variant of Huntington Disease” OR “Akinetic Rigid Variant of Huntington Disease” OR “Dementia” OR “Delirium” OR “Amnesia” OR “Dementias” OR “Amentia” OR “Amentias” OR “Deliriums” OR “Tinnitus”[Mesh] OR “Otitis Media”[Mesh] OR “Tinnitus” OR “Middle Ear Inflammation” OR “ear buzzing” OR “false latent otitis” OR “middle ear infection” OR “middle ear infections” OR “tympanitis”) NOT (“animals” [Mesh] NOT “humans”[Mesh])

Limits: English

C&P Embase (80)

‘Deafness’ OR ‘Deaf’ OR ‘Bilateral Deafness’ OR ‘Complete Hearing Loss’ OR ‘Prelingual Deafness’ OR ‘Deaf-Mutism’ OR ‘Deaf Mutism’ OR ‘Acquired Deafness’ OR ‘Extreme Hearing Loss’ AND (‘pediatrics’/exp OR ‘child’/exp OR ‘adolescent’/exp OR ‘Child’ OR ‘Children’ OR ‘Children’ OR ‘Disabled Child’ OR ‘Handicapped Children’ OR ‘Children with Disability’ OR ‘Children with Disabilities’ OR ‘Preschool Child’ OR ‘Preschool Children’ OR ‘Adolescent’ OR ‘Adolescents’ OR Teen* OR ‘Youth’ OR ‘Youths’ OR ‘Adolescence’ OR ‘girl’ OR ‘girls’ OR ‘boy’ OR ‘boys’ OR ‘juvenile’ OR ‘juveniles’ OR ‘Pediatrics’ OR ‘pediatric’ OR ‘pediatry’ OR ‘section 7’) AND (‘quality of life’/exp OR ‘Quality of Life’ OR ‘Life Qualities’ OR ‘Life Quality’ OR ‘HRQL’ OR ‘health related quality of life’ OR ‘HRQOL’) AND (‘instrumentation’/exp OR ‘validation study’/exp OR ‘reproducibility’/exp OR reproducib* OR ‘psychometry’/exp OR psychometr*OR clinimetr*OR clinometr* OR ‘observer variation’/exp OR ‘observer variation’ OR ‘discriminant analysis’/exp OR reliab* OR valid* OR coefficient OR ‘internal consistency’ OR ‘item correlation’ OR ‘item correlations’ OR ‘item selection’ OR ‘item selections’ OR ‘item reduction’ OR ‘item reductions’ OR agreement OR precision OR imprecision OR ‘precise values’ OR test–retest OR stability OR interrater OR inter-rater OR intrarater OR intra-rater OR intertester OR inter-tester OR intratester OR intra-tester OR interobserver OR inter-observer OR intraobserver OR intra-observer OR intertechnician OR intertechnician OR intratechnician OR intra-technician OR interexaminer OR inter-examiner OR intraexaminer OR intra-examiner OR interassay OR inter-assay OR intraassay OR intra-assay OR interindividual OR inter-individual OR intraindividual OR intra-individual OR interparticipant OR inter-participant OR intraparticipant OR intra-participant OR kappa OR kappas OR ‘coefficient of variation’ OR repeatab* OR generaliza* OR generalisa* OR concordance OR discriminative OR ‘known group’ OR ‘factor analysis’ OR ‘factor analyses’ OR ‘factor structure’ OR ‘factor structures’ OR dimensionality OR subscale* OR ‘multitrait scaling analysis’ OR ‘multitrait scaling analyses’ OR ‘item discriminant’ OR ‘interscale correlation’ OR ‘interscale correlations’ OR ‘individual variability’ OR ‘interval variability’ OR ‘rate variability’ OR ‘variability analysis’ OR ‘standard error of measurement’ OR sensitiv* OR responsive* OR ‘minimal detectable concentration’ OR interpreta* OR ‘meaningful change’ OR ‘minimal important change’ OR ‘minimal important difference’ OR ‘minimally important change’ OR ‘minimally important

difference’ OR ‘minimal detectable change’ OR ‘minimal detectable difference’ OR ‘minimally detectable change’ OR ‘minimally detectable difference’ OR ‘minimal real change’ OR ‘minimal real difference’ OR ‘minimally real change’ OR ‘minimally real difference’ OR ‘ceiling effect’ OR ‘floor effect’ OR ‘Item response model’ OR IRT OR Rasch OR ‘Differential item functioning’ OR DIF OR ‘computer adaptive testing’ OR ‘item bank’ OR ‘cross cultural equivalence’) NOT (‘cognition disorder’ OR ‘cognition disorders’ OR ‘cognitive defects’ OR ‘cognitive deficit’ OR ‘cognitive disability’ OR ‘cognitive disorder’ OR ‘cognitive disorders’ OR ‘cognitive dysfunction’ OR ‘cognitive impairment’ OR ‘delirium’ OR ‘dementia’ OR ‘amnestic’ OR ‘cognitive disorders’ OR ‘overinclusion’ OR ‘response interference’ OR ‘Cognition Disorders’ OR ‘Cognition Disorders’ OR ‘Auditory Perceptual Disorder’ OR ‘Psychoacoustical Disorders’ OR ‘Psychoacoustical Disorder’ OR ‘Auditory Comprehension Disorder’ OR ‘Auditory Comprehension Disorders’ OR ‘Auditory Processing Disorder’ OR ‘Auditory Processing Disorders’ OR ‘Acoustic Perceptual Disorder’ OR ‘Acoustic Perceptual Disorders’ OR ‘Auditory Inattention’ OR ‘Auditory Inattentions’ OR ‘Huntington Disease’ OR ‘Huntington Chorea’ OR ‘Huntingtons Disease’ OR ‘Huntington Chronic Progressive Hereditary Chorea’ OR ‘Chronic Progressive Hereditary Chorea’ OR ‘Huntingtons Chorea’ OR ‘Late-Onset Huntington Disease’ OR ‘Late Onset Huntington Disease’ OR ‘Juvenile Huntington Disease’ OR ‘Juvenile-Onset Huntington Disease’ OR ‘Juvenile Onset Huntington Disease’ OR ‘Akinetic-Rigid Variant of Huntington Disease’ OR ‘Akinetic Rigid Variant of Huntington Disease’ OR ‘Dementia’ OR ‘Delirium’ OR ‘Amnesia’ OR ‘Dementias’ OR ‘Amentia’ OR ‘Amentias’ OR ‘Deliriums’ OR ‘tinnitus’/exp OR ‘otitis media’/exp OR ‘tinnitus’ OR ‘Middle Ear Inflammation’ OR ‘ear buzzing’ OR ‘false latent otitis’ OR ‘middle ear infection’ OR ‘middle ear infections’ OR ‘tympanitis’) NOT ([animals]/lim NOT [humans]/lim) AND [english]/lim

C&P Scopus (181)

TITLE-ABS-KEY(“Deafness” OR “Deaf” OR “Bilateral Deafness” OR “Complete Hearing Loss” OR “Prelingual Deafness” OR “Deaf Mutism” OR “Acquired Deafness” OR “Extreme Hearing Loss”)

AND

TITLE-ABS-KEY(“pediatrics” OR “child” OR “adolescent” OR “Child” OR “Children” OR “Children” OR “Disabled Child” OR “Handicapped Children” OR “Children with Disability” OR “Children with Disabilities” OR “Preschool Child” OR “Preschool Children” OR “Adolescent” OR “Adolescents” OR Teen* OR “Youth” OR “Youths” OR “Adolescence” OR “girl” OR “girls” OR “boy” OR “boys” OR “juvenile” OR “juveniles” OR “Pediatrics” OR “pediatric” OR “pediatry” OR “section 7”)

AND

TITLE-ABS-KEY(“quality of life” OR “Quality of Life” OR “Life Qualities” OR “Life Quality” OR “HRQL” OR “health related quality of life” OR “HRQOL”)

AND

(“instrumentation” OR “validation study” OR “reproducibility” OR reproducibility OR “psychometry” OR “observer variation” OR “observer variation” OR “discriminant analysis” OR reliability OR validity OR coefficient OR “internal consistency” OR “item correlation” OR “item correlations” OR “item selection” OR “item selections” OR “item reduction” OR “item reductions” OR agreement OR precision OR imprecision OR “precise values” OR test retest OR stability OR interrater OR interrater OR intrarater OR intrarater OR intertester OR intertester OR intratester OR intratester OR interobserver OR interobserver OR intraobserver OR intraobserver OR intertechnician OR intertechnician OR intratechnician OR intratechnician OR interexaminer OR interexaminer OR intraexaminer OR intraexaminer OR interassay OR interassay OR intraassay OR intraassay OR interindividual OR interindividual OR intraindividual OR intraindividual OR interparticipant OR interparticipant OR intraparticipant OR intraparticipant OR kappa OR kappas OR “coefficient of variation” OR repeatability OR generalizability OR generalisation OR concordance OR discriminative OR “known group” OR “factor analysis” OR “factor analyses” OR “factor structure” OR “factor structures” OR dimensionality OR subscale OR subscales OR “multitrait scaling analysis” OR “multitrait scaling analyses” OR “item discriminant” OR “interscale correlation” OR “interscale correlations” OR “individual variability” OR “interval variability” OR “rate variability” OR “variability analysis” OR “standard error of measurement” OR sensitivity OR responsiveness OR “minimal detectable concentration” OR interpretability OR “meaningful change” OR “minimal important change” OR “minimal important difference” OR “minimally important change” OR “minimally important difference” OR “minimal detectable change” OR “minimal detectable difference” OR “minimally detectable change” OR “minimally detectable difference” OR “minimal real change” OR “minimal real difference” OR “minimally real change” OR “minimally real difference” OR “ceiling effect” OR “floor effect” OR “Item response model” OR IRT OR Rasch OR “Differential item functioning” OR DIF OR “computer adaptive testing” OR “item bank” OR “cross cultural equivalence”

NOT

“cognition disorder” OR “cognition disorders” OR “cognitive defects” OR “cognitive deficit” OR “cognitive disability” OR “cognitive disorder” OR “cognitive disorders” OR “cognitive dysfunction” OR “cognitive impairment” OR “delirium” OR “dementia” OR “amnestic” OR “cognitive disorders” OR “overinclusion” OR “response interference” OR “Cognition Disorders” OR “Cognition Disorders” OR “Auditory Perceptual Disorder” OR “Psychoacoustical Disorders” OR “Psychoacoustical Disorder” OR “Auditory Comprehension Disorder” OR “Auditory Comprehension Disorders” OR “Auditory Processing Disorder” OR “Auditory Processing Disorders” OR “Acoustic Perceptual Disorder” OR “Acoustic Perceptual Disorders” OR “Auditory Inattention” OR “Auditory Inattentions” OR “Huntington Disease” OR “Huntington Chorea” OR “Huntingtons Disease” OR “Huntington Chronic Progressive Hereditary Chorea” OR “Chronic Progressive Hereditary Chorea” OR “Huntingtons Chorea” OR “Late-Onset Huntington Disease” OR “Late Onset Huntington Disease” OR “Juvenile Huntington Disease” OR “Juvenile-Onset Huntington Disease” OR “Juvenile Onset Huntington Disease” OR “Akinetic-Rigid Variant of Huntington Disease” OR “Akinetic Rigid Variant of Huntington

Disease” OR “Dementia” OR “Delirium” OR “Amnesia” OR “Dementias” OR “Amentia” OR “Amentias” OR “Deliriums” OR “tinnitus” OR “otitis media” OR “tinnitus” OR “Middle Ear Inflammation” OR “ear buzzing” OR “false latent otitis” OR “middle ear infection” OR “middle ear infections” OR “tympanitis”d

Limited to: (Human OR Humans) AND English

C&P CINAHL (13)

(MH “Deafness+” OR “Deafness” OR “Deaf” OR “Bilateral Deafness” OR “Complete Hearing Loss” OR “Prelingual Deafness” OR “Deaf-Mutism” OR “Deaf Mutism” OR “Acquired Deafness” OR “Extreme Hearing Loss”) AND (MH “Child+” OR MH “Adolescence+” OR “Minors” OR “Preschool Child” OR “Preschool Children” OR “Child” OR “Children” OR “Disabled Child” OR “Handicapped Children” OR “Children with Disability” OR “Children with Disabilities” OR “Adolescent” OR “Adolescents” OR “Teens” OR “Teen” OR “Teenagers” OR “Teenager” OR “Youth” OR “Youths” OR “Adolescence” OR “girl” OR “girls” OR “boy” OR “boys” OR “juvenile” OR “juveniles”) AND (MH “Quality of Life” OR “quality of life” OR “Life Qualities” OR “Life Quality” OR “HRQL” OR “health related quality of life” OR “HRQOL”) AND (MH “Validation Studies” OR MH “Reproducibility of Results” OR MH “Psychometrics” OR psychometr*OR clinimetr*OR clinometr* OR “observer variation” OR “observer variation” OR “discriminant analysis” OR reliab* OR valid* OR coefficient OR “internal consistency” OR “item correlation” OR “item correlations” OR “item selection” OR “item selections” OR “item reduction” OR “item reductions” OR agreement OR precision OR imprecision OR “precise values” OR test–retest OR stability OR interrater OR inter-rater OR intrarater OR intra-rater OR intertester OR inter-tester OR intratester OR intra-tester OR interobserver OR inter-observer OR intraobserver OR intra-observer OR intertechnician OR intertechnician OR intratechnician OR intra-technician OR interexaminer OR inter-examiner OR intraexaminer OR intra-examiner OR interassay OR inter-assay OR intraassay OR intra-assay OR interindividual OR inter-individual OR intraindividual OR intra-individual OR interparticipant OR inter-participant OR intraparticipant OR intra-participant OR kappa OR kappas OR “coefficient of variation” OR repeatab* OR generaliza* OR generalisa* OR concordance OR discriminative OR “known group” OR “factor analysis” OR “factor analyses” OR “factor structure” OR “factor structures” OR dimensionality OR subscale* OR “multitrait scaling analysis” OR “multitrait scaling analyses” OR “item discriminant” OR “interscale correlation” OR “interscale correlations” OR “individual variability” OR “interval variability” OR “rate variability” OR “variability analysis” OR “standard error of measurement” OR sensitiv* OR responsive* OR “minimal detectable concentration” OR interpreta* OR “meaningful change” OR “minimal important change” OR “minimal important difference” OR “minimally important change” OR “minimally important difference” OR “minimal detectable change” OR “minimal detectable difference” OR “minimally detectable change” OR “minimally detectable difference” OR “minimal real change” OR “minimal real difference” OR “minimally real change” OR “minimally real difference” OR “ceiling effect” OR “floor effect” OR “Item response model” OR IRT OR Rasch OR “Differential item functioning” OR DIF OR “computer adaptive testing” OR “item bank” OR “cross cultural equivalence”) NOT (“cognition disorder” OR “cognition

disorders” OR “cognitive defects” OR “cognitive deficit” OR “cognitive disability” OR “cognitive disorder” OR “cognitive disorders” OR “cognitive dysfunction” OR “cognitive impairment” OR “delirium” OR “dementia” OR “amnestic” OR “cognitive disorders” OR “overinclusion” OR “response interference” OR “Cognition Disorders” OR “Cognition Disorders” OR “Auditory Perceptual Disorder” OR “Psychoacoustical Disorders” OR “Psychoacoustical Disorder” OR “Auditory Comprehension Disorder” OR “Auditory Comprehension Disorders” OR “Auditory Processing Disorder” OR “Auditory Processing Disorders” OR “Acoustic Perceptual Disorder” OR “Acoustic Perceptual Disorders” OR “Auditory Inattention” OR “Auditory Inattentions” OR “Huntington Disease” OR “Huntington Chorea” OR “Huntington”s Disease” OR “Huntington Chronic Progressive Hereditary Chorea” OR “Chronic Progressive Hereditary Chorea” OR “Huntington”s Chorea” OR “Late-Onset Huntington Disease” OR “Late Onset Huntington Disease” OR “Juvenile Huntington Disease” OR “Juvenile-Onset Huntington Disease” OR “Juvenile Onset Huntington Disease” OR “Akinetic-Rigid Variant of Huntington Disease” OR “Akinetic Rigid Variant of Huntington Disease” OR “Dementia”OR “Delirium” OR “Amnesia” OR “Dementias” OR “Amentia” OR “Amentias” OR “Deliriums” OR MH “Tinnitus” OR MH “Otitis Media” OR “Tinnitus” OR “Middle Ear Inflammation” OR “ear buzzing” OR “false latent otitis” OR “middle ear infection” OR “middle ear infections” OR “tympanitis”)

Grey Literature Search

Proquest Dissertations and Theses—TI,AB(Deaf OR deafness OR “Complete Hearing Loss” OR “Prelingual Deafness” OR “Deaf-Mutism” OR “Deaf Mutism” OR “Extreme Hearing Loss”) AND TI,AB(child OR children OR infant* OR adolescen* OR pediatrics OR teen* OR youth OR youths OR girl* OR boy* OR juvenile*) AND TI,AB(“Quality of Life” OR “life qualities” OR “life quality” OR “HRQL” OR “health related quality of life” OR “HRQOL”)

FirstSearch Proceedings—(kw: deaf OR kw: deafness OR kw: complete w hearing w loss OR kw: prelingual w deafness OR kw: deaf-mutism OR kw: deaf w mutism OR kw: extreme w hearing w loss) AND (kw: child OR kw: children OR kw: infant OR kw: adolescent OR kw: pediatric OR kw: youth OR kw: girls OR kw: boys OR kw: juvenile OR kw: juveniles) AND (kw: quality w1 life OR kw: life w qualities OR kw: life w quality OR kw: HRQL OR kw: health w related w quality w1 life OR kw: HRQOL) and ln= “english”

ClinicalTrials.Gov—(Deaf OR deafness) AND (child OR children OR infant* OR adolescen* OR pediatrics OR teen OR juvenile*) AND (“Quality of Life” OR “life qualities” OR “life quality” OR “HRQL” OR “health related quality of life” OR “HRQOL”)

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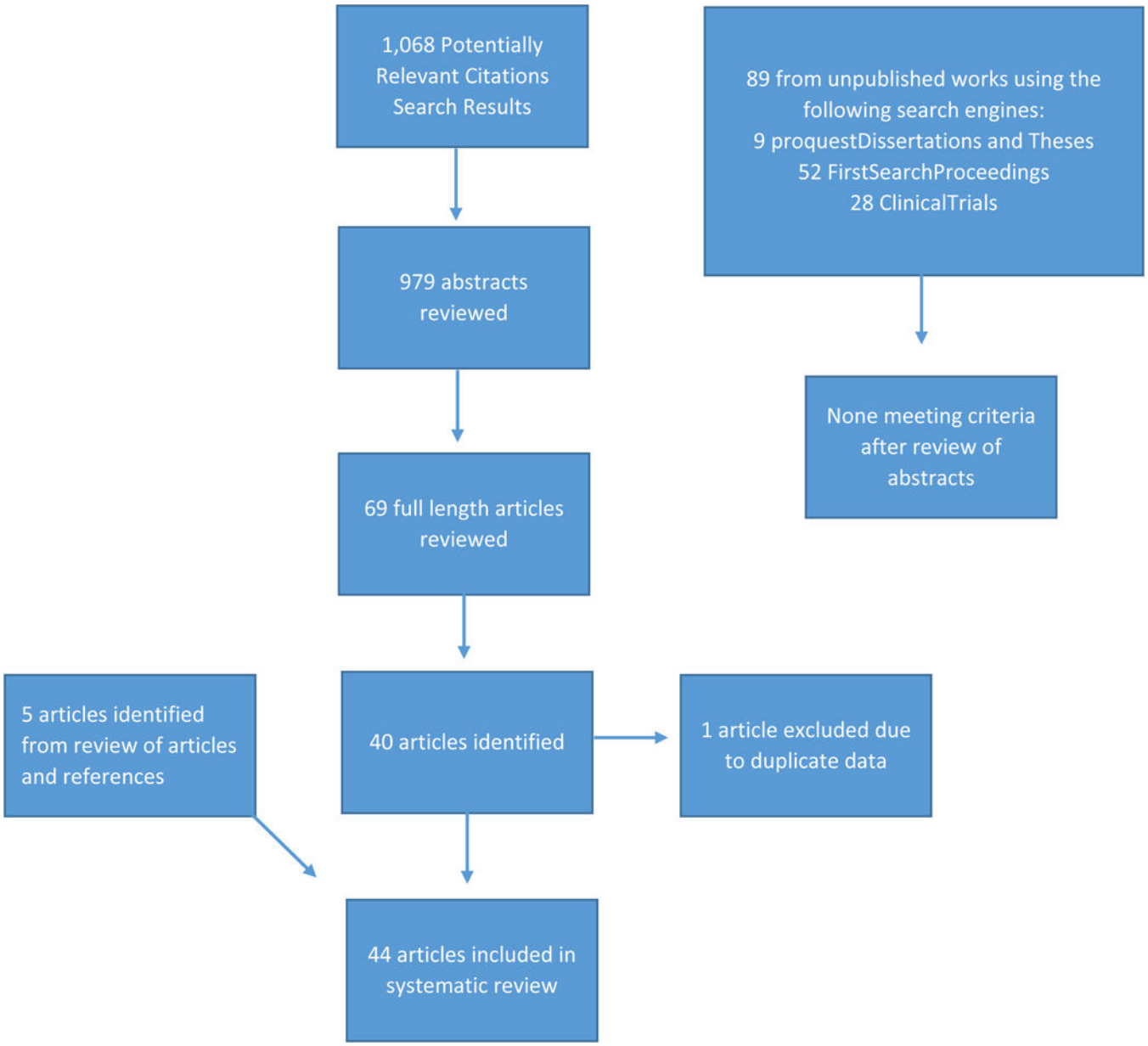

Figure 1.
Flow chart of systematic review

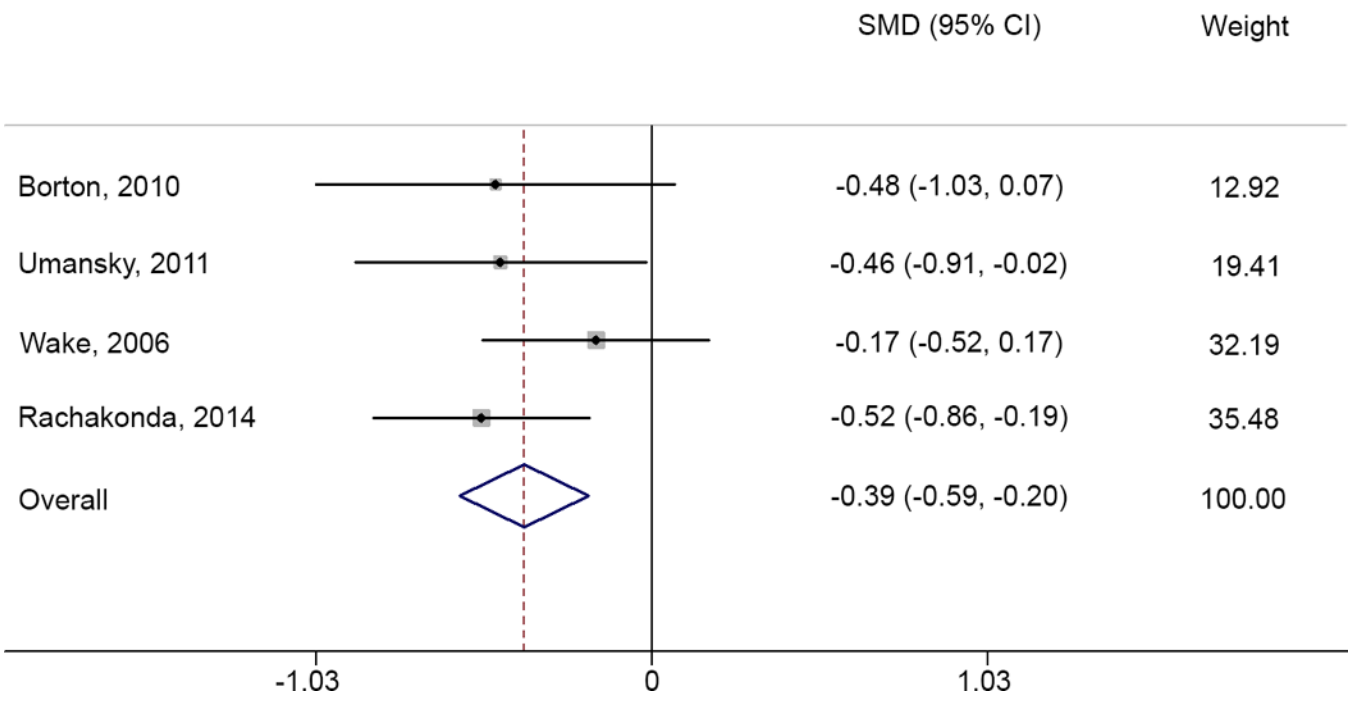

Figure 2.
PedsQL – School Domain: comparing normal hearing (NH) to bilateral hearing loss (BHL)

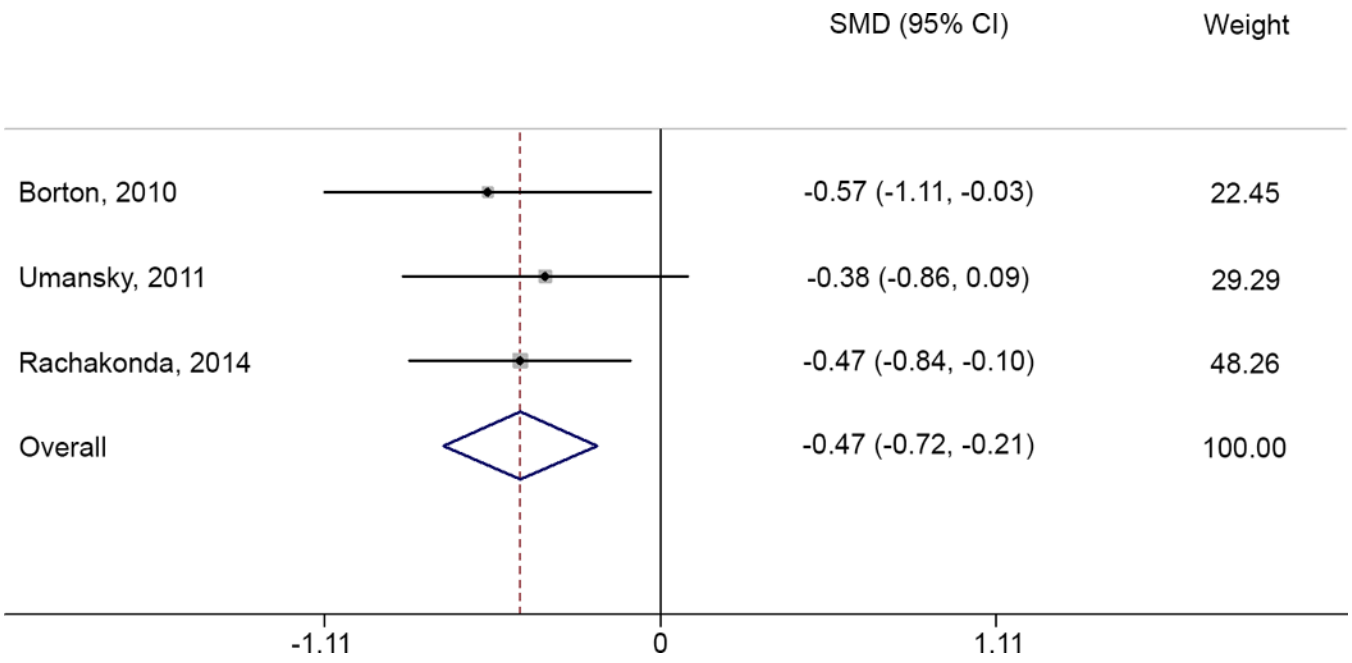

Figure 3.
PedsQL – School Domain: comparing normal hearing (NH) to unilateral hearing loss (UHL)

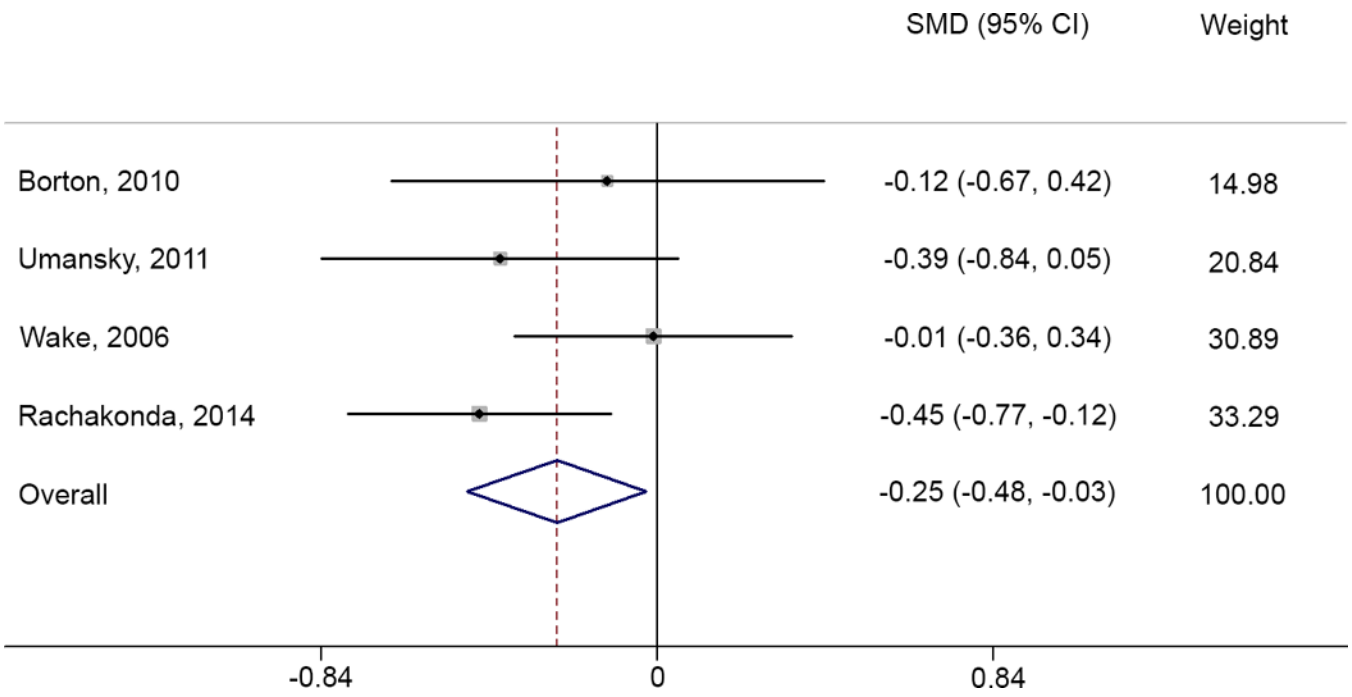

Figure 4.
PedsQL – Social Domain: comparing normal hearing (NH) to bilateral hearing loss (BHL)

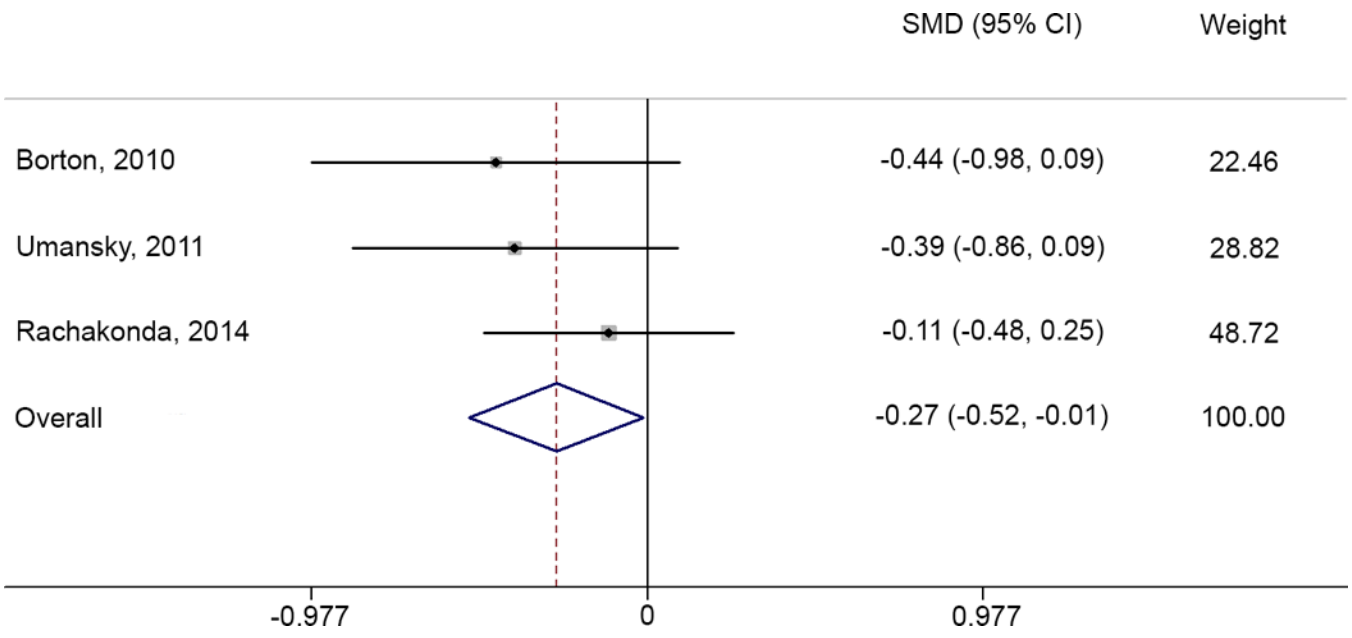

Figure 5.
PedsQL – Social Domain: comparing normal hearing (NH) to unilateral hearing loss (UHL)

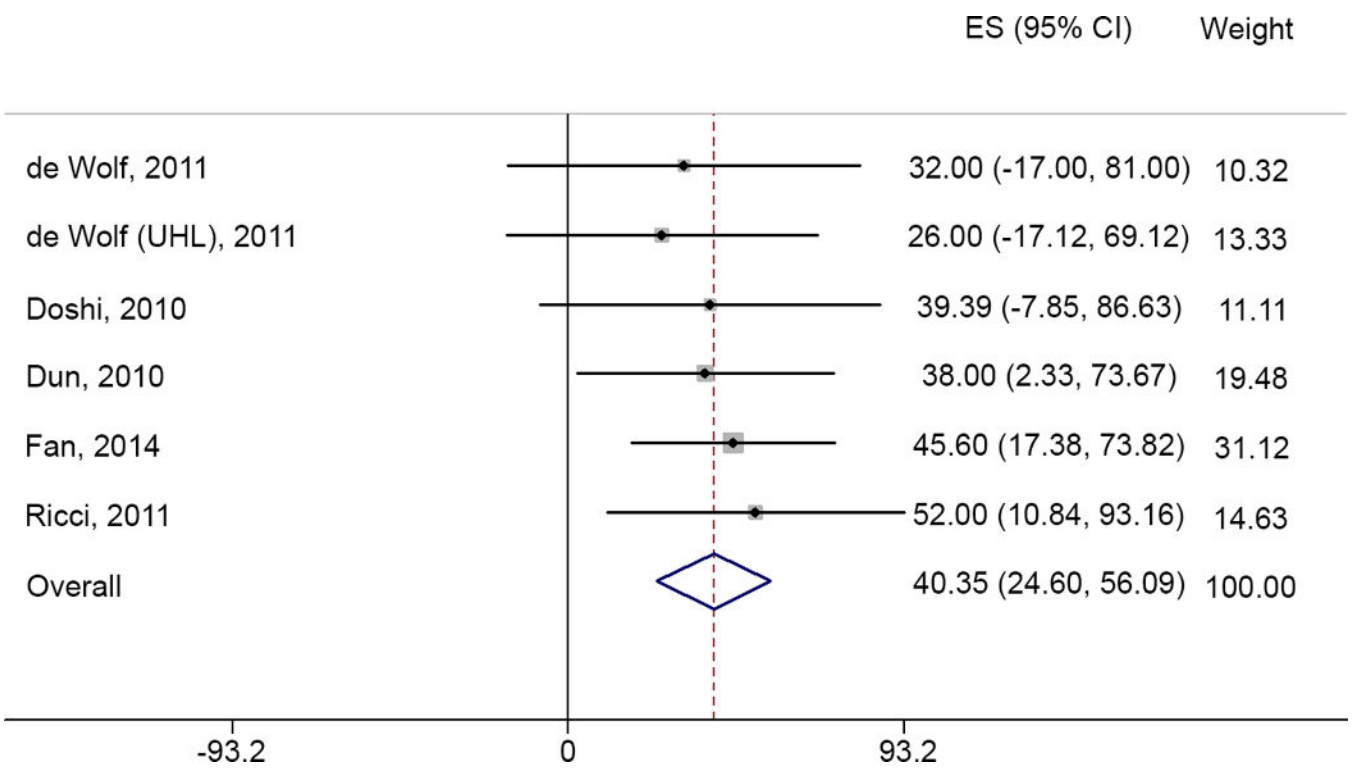

Figure 6.
GCBI Improvement following BAHA placement – all children with hearing loss

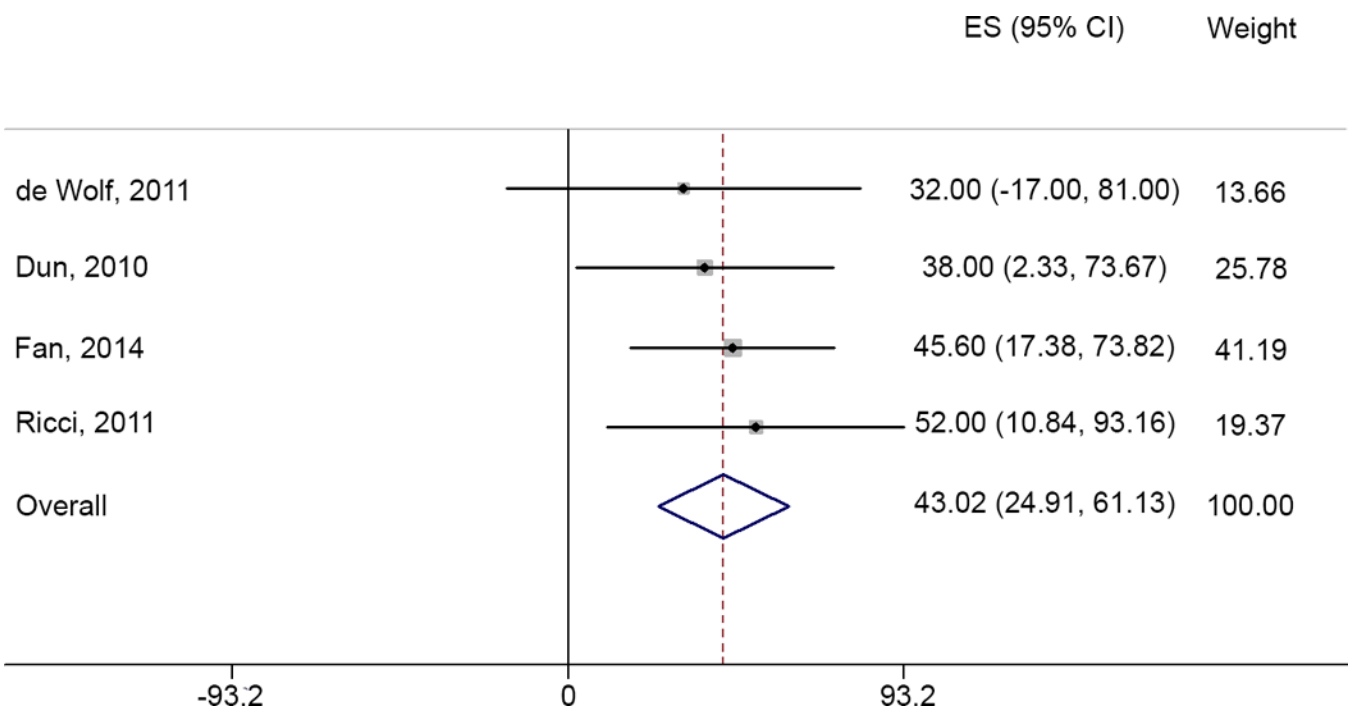

Figure 7.
GCBI Improvement following BAHA placement – bilateral hearing loss only

Table 1

Quality Assessment

Year	Author	Selection bias	Outcome bias	Overall risk of bias
2010	Borton, et al[24]	Low	Low	Low
2013	Elbasan, et al[25]	Low	Low	Low
2008	Fellinger, et al[26]	Unclear	Low	Unclear
2005	Huber, et al[27]	Unclear	Low	Unclear
2010	Lovett, et al[28]	Unclear	Low	Unclear
2010	Loy, et al[29]	Unclear	Low	Unclear
2007	Petrou, et al[30]	Low	Low	Low
2012	Schick, et al[31]	Low	Low	Low
2011	Umansky, et al[22]	Low	Low	Low
2004	Wake, et al[32]	Low	Low	Low
2004	Wake, et al (b)[33]	Low	Low	Low
2006	Wake, et al[34]	Low	Low	Low
2009	Warner-czyz, et al[35]	Unclear	Low	Unclear
2014	Rachakonda et al[23]	Low	Low	Low
2011	Hintermair, et al[36]	Unclear	Low	Unclear
2012	Clark, et al[37]	Low	Low	Low
2013	Banga, et al[18]	Unclear	Low	Unclear
2011	de Wolf, et al[17]	Low	Low	Low
2013	Doshi, et al[16]	Low	Low	Unclear
2010	Dun, et al[13]	Low	Low	Low
2014	Fan, et al[14]	Low	Low	Low
2008	Kunst, et al[19]	Low	Low	Low
2011	Ricci, et al[38]	Low	Low	Low
2006	Barton, et al[39]	Unclear	Low	Unclear
2006	Stacey, et al[40]	Low	Unclear	Unclear
2000	Cheng, et al[41]	Low	Low	Low
2009	Huttunen, et al[42]	Low	Low	Low
2007	Sach, et al[43]	Unclear	Low	Unclear
2009	Schorr, et al[44]	Low	Low	Low
2012	Necula, et al[45]	Low	Low	Low
2011	Briggs, et al[46]	Low	Low	Low
2012	Sparreboom, et al[47]	Low	Low	Low
2007	Beijen, et al[48]	Low	Low	Low
2007	Keilmann, et al[49]	Unclear	Low	Unclear
2010	Korver, et al[50]	Low	Low	Low

Year	Author	Selection bias	Outcome bias	Overall risk of bias
2011	Kushalnagar, et al[51]	Unclear	Low	Unclear
2013	Meyer, et al[52]	Unclear	Low	Unclear
2011	Patrick, et al[53]	Unclear	Low	Unclear
2008	Smith-Olinde, et al[54]	Low	Low	Low
2011	Warner-Czyz, et al[55]	Unclear	Low	Unclear
2010	Rajendran, et al[56]	Unclear	Low	Unclear

Table 2

Quality of life assessments: pediatric patients with hearing loss as compared to normal hearing peers

Year	Authors	Age (years)	NH (n)	HL (n)	QOL tools	Conclusions
2010	Borton, et al.[57]	6–17	25	61	PedsQL	No statistical difference in quality of life between children with normal vs impaired hearing (total, psychosocial, physical)
2013	Elbasan, et al.[25]	5–7	28	27	CHQ, PEDI	Children with HL as compared to normal hearing were found to have lower scores in the following domains: global general health, emotional, physical, behavior, mental health, general health, family activity and cohesion and parental time (CHQ-PF50) and self-care, mobility and social function (PEDI). ($p<0.05$ for all listed comparisons above.)
2008	Fellinger, et al.[26]	6–16	0	99	Inventory of Life Quality	As compared to a normative sample of normal hearing children, deaf pupils reported worse quality of life. ($p<0.05$ for the following comparisons: emotional problems, conduct problems, peer problems, social behavior and hyperactivity.)
2005	Huber, et al.[27]	8–16	1501	29	KINDLR	As compared to normalized scores, children with CI had lower quality of life (ES: −1.6, $p<0.001$ for females with CI: age 8–12, self and ES: −1.1, $p<0.025$ for males with CI: age 8–12, self).
2010	Lovett, et al.[28]	1–16	56	50	HUI3	There were no significant differences between the 3 groups, normal hearing, unilateral CI and bilateral CI in health utility or QOL.
2010	Loy, et al.[29]	11–16	1501	88	KINDLR	The two participant groups, those with Cis for hearing loss and their normal hearing peers, did not differ in their reported overall quality of life.
2007	Petrou, et al.[30]	7–9	63	120	HUI3	As compared to their normal hearing peers, children with bilateral permanent hearing loss were found to have significantly lower scores in the following domains of HUI3: vision, hearing, speech, ambulation, dexterity and cognition ($p<0.05$ for all comparisons listed).
2012	Schick, et al.[31]	11–18	0	221	YQOL-R, YQOL-DHH	When compared to scores for their normal hearing peers, participants who were deaf or hard of hearing scored lower in “self” ($p=0.036$) and “relationship” domains ($p=0.003$).
2011	Umansky, et al.[22]	7–12	35	80	PedsQL, HEAR-QL	Children with hearing loss reported significantly lower scores than their normal hearing peers on the HEAR-QL (71[18] vs 98[5]; $p<0.001$). PedsQL scores were not statistically different between hearing loss and normal hearing patients.
2004	Wake, et al.[32]	7–8	895	83	CHQ	This study compared children with hearing loss fitted for hearing aids or CI compared to their normal hearing peers. Parent reported psychosocial well-being was lower in children with hearing loss ($p = 0.001$).
2004	Wake, et al.[33]	7–8	865	86	CHQ	Compared to a normative sample, children with hearing loss had statistically significantly lower scores in quality of life measures related to psychosocial skills than their normal hearing peers (ES: 0.5, $p<0.001$). There were no differences in the physical domain.
2006	Wake, et al.[34]	not given	6516	55	PedsQL	Compared to a normative sample of normal hearing peers, there were no significant differences in quality of life in hearing impaired children.
2009	Warner-czyz, et al[35]	4–7	25	50	KINDLR	This study compared patients with hearing impairment and CI and those with normal hearing. There were no differences found in quality of life between these two groups.
2014	Rachakonda et al.[23]	13–18	54	172	PedsQL, HEAR-QL	As compared to normal hearing peers, children with hearing loss reported significantly poorer hearing related QOL on the HEAR-QL in all categories ($p<0.001$) as well as lower scores on the school domain of the PedsQL (79[17] vs 70[20] $p=0.001$).

Year	Authors	Age (years)	NH (n)	HL (n)	QOL tools	Conclusions
2011	Hintermair, et al.[36]	6–18	0	212	Inventory of Life Quality	Compared to a normative sample, those with hearing loss reported worse quality of life in school ($p<0.001$) and social domains ($p<0.001$) than their normal hearing peers.
2012	Clark, et al.[37]	0–5	97	188	VAS-HRQoL	Baseline deficits of CI candidates as compared to normal hearing peers were found in development domains, cognition (ES: 1.4, $p=0.01$) and speech recognition (ES: 1.4, $p=0.02$). Children implanted at a younger age had greater improvement in development.
2010	Rajendran, et al.[56]	6–11	100	100	PedsQL	Compared to age-matched controls, children with hearing loss had lower scores in the physical and social domains. Specific domain scores and effect size were not available from this paper and it was not included in the meta-analysis.

Table 3

Quality of life assessments following intervention for hearing impairment

Year	Authors	Age (years)	HL (n)	QOL tools	Intervention	Conclusions
2013	Banga, et al.[18]	6–16	27	GCBI	BAHA	26 of 27 children reported an improvement on GCBI after surgery.
2011	de Wolf, et al.[17]	6–17	31	GCBI, HUI3, APHAB	BAHA	Patients reported an improvement in GCGI (+32[25] for bilateral hearing loss and +26[22] for unilateral hearing loss) following use of their BAHAs.
2013	Doshi, et al.[16]	7–12	8	GCBI, SSQ	BAHA	7 of the 8 children showed an increase in GCGI score (+40[24]) after surgery and 5/8 children felt that their quality of life had improved.
2010	Dun, et al.[13]	5–28	21	GCBI	BAHA	A benefit from using the BAHA implant was found using the GCGI (+38[18]), with the learning domain showing the most positive change.
2014	Fan, et al.[14]	6–28	16	GCBI	BAHA	There was an improvement in all GCBI scores following surgery (+46[14]).
2008	Kunst, et al.[19]	5–18	10	GCBI, SSQ	BAHA	GCBI scores increased following BAHA implantation (+34).
2011	Ricci, et al.[38]	5–14	31	GCBI	BAHA	Quality of life improved for all patients who underwent BAHA (+52[21]).
2006	Barton, et al.[39]	not given	2266	HUI3	CI	This study compared patients who were implanted as compared to those with hearing-impairment who were not implanted. There was a significant health utility benefit of implantation, especially with young age at implantation.
2006	Stacey, et al.[40]	3–20	2858	Quality of life questionnaire	CI	Parents and teachers completed questionnaires. Pediatric CI at less than 5 years of age was associated with improvement in communication, education and quality of life. Of note, this study used a validated QOL measure, but then modified it before use.
2000	Cheng, et al[41]	not given	22	HUI3	CI	Following implantation, 95% of HUI3 scores increased (increase from 0.25 to 0.39), this finding was due to improvements in hearing and speech domains.
2009	Huttunen, et al.[42]	1–12	36	Children with cochelar implants: parental perspectives	CI	Following implantation, parents reported improvement in social relations, communication and general functioning.
2007	Sach, et al.[43]	not given	222	EuroQoL	CI	Quality of life was not significantly improved following implantation.
2009	Schorr, et al.[44]	5–14	37	Children's quality of life questionnaire	CI	Children reported an improvement in quality of life from their CI (mean of 26 points out of 35).
2012	Necula, et al.[45]	2–12	134	Nijmegen CI HRQoL	CI/hearing-aid	CI improved auditory performance and speech production much more than hearing aids ($p<0.05$ in all domains).
2011	Briggs, et al.[46]	7–12	8	HEAR-QL	Hearing aids	There was an improvement in quality of life following hearing aid use (difference of 20.1[10.5], p=0.002)
2012	Sparreboom, et al.[47]	1–7	39	HUI3, PedsQL, GCBI, SSQ, NCIQ	second CI	Generic quality of life measures did not detect any differences following second implant. Higher scores were found for bilaterally implanted children in all domains of the GCBI as well as the SSQ and in sound perception, speech production and activity domains of the NCIQ. Bilaterally implanted children showed continued improvement in QoL over time, as compared to unilaterally implanted patients.
2007	Beijen, et al.[48]	1–2	10	SSQ, PedsQL	second CI	This study compared bilaterally implanted children to unilaterally implanted children. No significant differences were found between SSQ and PedsQL scores.

Table 4

Quality of life in hearing impaired children: Miscellaneous studies evaluating children with hearing impairment

Year	Authors	Age (years)	HL (n)	QOL tools	Conclusions
2007	Keilmann, et al.[49]	6–11	131	Frankfurt Self Concept Scales for Children	This study evaluated children placed in assistive schools as compared to mainstream schools. Physical well-being was not affected, however children with hearing impairment placed in special schools had lower confidence and worse quality of life than those with hearing impairment who were in mainstream schools ($p<0.05$).
2010	Korver, et al.[50]	3–5	301	PedsQL	This study evaluated children who underwent newborn screening as compared to screening at a later date. Quality of life was found to be statistically higher in the group of patients who underwent newborn hearing screening as compared to later assessment (mean between-group difference of 5.3(95% CI: 1.7–8.9)).
2011	Kushalnagar, et al.[51]	11–18	230	YQoL-R, YQOL-DHH	Multiple cofactors were assessed. Quality of life scores were associated with an ability to understand parental expression and communication.
2013	Meyer, et al.[52]	11–18	157	YQOL-DHH	Quality of life was assessed as related to multiple aspects of hearing impairment. Youth who were not using cochlear implants or technology scored higher on this stigma-related QoL ($p<0.05$) as compared to those with hearing aids or CI.
2011	Patrick, et al.[53]	11–18	230	YQOL-R, YQOL-DHH	Patients had a variable amount of hearing loss in this study, and approximately 1/4 had previously received CI. Interestingly, quality of life was not significantly associated with hearing level.
2008	Smith-Olinde, et al.[54]	5–10	103	HUI3 QWB	Parents reported that QOL correlated with pure-tone average and was positively associated with CI status.
2011	Warner-Czyz, et al.[55]	4–16	138	KINDL	All children in this study were CI users. Younger children rated HRQoL more positively than older children (mean 75.4[9.2] vs 70.4[12.4]).

Table 5

PedsQL Meta-Analysis

Comparison	Effect size (SE)	Difference in means (SE)	95% CI of point difference*		p-value*	Statistical significance	Clinical significance
Total NH and UHL	−0.27 (0.13)	−3.80 (1.84)	−7.40	−0.20	0.038	√	
Total NH and BHL	−0.26 (0.11)	−3.41 (1.76)	−6.86	0.05	0.053		
Physical NH and UHL	−0.11 (0.13)	−1.52 (1.78)	−5.01	1.98	0.395		
Physical NH and BHL	−0.21 (0.10)	−3.15 (1.49)	−6.07	−0.23	0.034	√	
Emotional NH and UHL	−0.18 (0.13)	−3.53 (2.53)	−8.49	1.43	0.163		
Emotional NH and BHL	−0.13 (0.10)	−2.61 (1.96)	−6.44	1.23	0.183		
Social NH and UHL	−0.27 (0.13)	−4.74 (2.29)	−9.22	−0.26	0.038	√	√
Social NH and BHL	−0.25 (0.11)	−4.31 (2.16)	−8.54	−0.07	0.046	√	√
School NH and UHL	−0.47 (0.13)	−8.79 (2.43)	−13.55	−4.03	<0.001	√	√
School NH and BHL	−0.39 (0.10)	−6.93 (1.77)	−10.40	−3.47	<0.001	√	√

Variance, 95% CI and p-value based on difference in means.

NH = normal hearing, UHL = unilateral hearing loss, BHL = bilateral hearing loss