Hearing-related quality of life, developmental outcomes and performance in children and young adults with unilateral conductive hearing loss due to aural atresia.

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List of abbreviations and relevant definitions

ASR	Adult Self-Report
CBCL	Children's Behaviour Checklist
CCC-2-NL	Dutch version of the Children's Communication Checklist
dB	Decibel
dB nHL	Decibel above normal hearing level
iqr	Interquartile range
M	Mean
sd	Standard deviation
SNHL	Sensorineural Hearing Loss
SPSS	Statistical Package for the Social Sciences
SSQ	Speech, Spatial and Quality of Hearing Scale
YSR	Youth Self-Report

SUMMARY

Background: While research has shown that children with unilateral inner ear deafness have a lower quality of life and developmental outcomes compared to normal hearing peers, little is known about these domains in children with unilateral congenital conductive hearing loss due to aural atresia.

Aims: The aim of this study is to investigate the hearing-related quality of life and developmental outcomes and performance in children and young adults with unilateral conductive hearing loss due to aural atresia.

Method: Nineteen children and young adults with unilateral aural atresia received a set of five questionnaires. Hearing-related quality of life, general quality of life, speech and language development, educational performance and social-emotional development were measured by the Speech, Spatial and Quality of Hearing Scale, Kidscreen-27, Children's Communication Checklist, a custom questionnaire, Children's Behaviour Checklist and Youth/Adult Self-Report, respectively. Scores on the questionnaires were compared to their norm scores. Mann-Whitney U tests and independent t-tests were used to identify significant differences between age groups.

Results: Mean scores on the Speech, Spatial and Quality of Hearing Scale were M=6.78, M=5.00 and M=6.98 for the Speech, Spatial and Quality of Hearing subscales, respectively. Mean scores on the Kidscreen-27, Children's Communication Checklist, Children's Behaviour Checklist and Youth/Adult Self-Report fell within normal or non-clinical range. A higher need for educational assistance was observed.

Conclusion: Children and young adults with unilateral conductive hearing loss due to congenital aural atresia seem to have a lower hearing-related quality of life compared to normal-hearing peers and seem to need educational assistance. Regarding general quality of life, speech and language development and social-emotional development these children and young adults seem to develop normally.

Recommendation: Guidance for these children during education is prudent to allow them to thrive.

Key words: Aural atresia, hearing loss, children, quality of life, development

SAMENVATTING

Achtergrond: Onderzoek heeft aangetoond dat kinderen met een defect binnenoor een lagere kwaliteit van leven hebben en zich slechter ontwikkelen dan normaal horende kinderen. Echter, er is nog weinig bekend over de ontwikkeling van kinderen met eenzijdig conductief gehoorverlies door aangeboren aurale atresie.

Doel: Het doel van dit onderzoek is het onderzoeken van gehoorgerelateerde kwaliteit van leven en ontwikkeling in verscheidene domeinen van kinderen en jongvolwassenen met eenzijdig conductief gehoorverlies door aangeboren aurale atresie.

Methode: Negentien kinderen en jong volwassenen met eenzijdige aurale atresie hebben een set van vijf vragenlijsten ontvangen. Gehoorgerelaterde kwaliteit van leven, algemene kwaliteit van leven, spraak-taalontwikkeling, schoolgerelateerde aspecten en sociaal-emotionele ontwikkeling is onderzocht door middel van respectievelijk de Speech, Spatial and Quality of Hearing Scale, Kidscreen-27, Children's Communication Checklist, een zelfgemaakte vragenlijst, Children's Behaviour Checklist en Youth/Adult Self-Report. Scores op de vragenlijsten zijn vergeleken met hun normen. Mann-Whitney U toetsen en onafhankelijke t-toetsen zijn gebruikt om verschillen aan te tonen.

Resultaten: Gemiddelde scores op de Speech, Spatial and Quality of Hearing Scale waren M=6.78, M=5.00 en M=6.98 op de Speech, Spatial en Quality of Hearing subschalen, respectievelijk. Gemiddelde scores op de Kidscreen-27, Children's Communication Checklist, Children's Behaviour Checklist en Youth/Adult Self-Report vielen allemaal binnen de normale of niet klinische marges van de norm. Kinderen hadden vaak onderwijsondersteuning nodig.

Conclusie: Kinderen en jongvolwassenen met eenzijdig conductief gehoorverlies door aangeboren aurale atresie lijken een lagere gehoorgerelateerde kwaliteit van leven te hebben en lijken vaker ondersteuning in het onderwijs nodig te hebben dan normaal horende kinderen. Wat betreft algemene kwaliteit van leven, spraak-taalontwikkeling en sociaal-emotionele ontwikkeling lijken deze kinderen en jongvolwassenen zich normaal te ontwikkelen.

Aanbevelingen: Onderwijsbegeleiding voor deze kinderen is aanbevolen om deze kinderen de kans te geven zich even goed te ontwikkelen als normaal horende kinderen.

Kernwoorden: Aurale atresie, gehoorverlies, kinderen, kwaliteit van leven, ontwikkeling

1. Introduction

Congenital aural atresia is a birth defect that results in an underdeveloped external auditory canal, either unilaterally or bilaterally. In some cases, the middle ear is affected as well, including the tympanic membrane and the ossicles.[1] Congenital aural atresia has a prevalence of 1 in 10,000-20,000 births[2] and is often accompanied with microtia. Microtia results in a malformed outer ear, with different grades in deformity ranging from slightly altered to non-existent.[3] Due to the deformities in the middle and/or outer ear canal, which are mostly unilateral, a conductive hearing loss is seen on the affected side.

Unilateral sensorineural severe to profound hearing loss (SNHL) gives major theoretical disadvantages in terms of speech perception in noise and localization of sounds compared to a binaural situation.[4] This results in functional disabilities with language delays in children as a consequence.[5] Regarding educational performance, unilateral SNHL results in increased rates of grade failure, need for speech therapy and need for additional educational assistance.[5–7] In addition, children with unilateral SNHL have been discovered to have a lower quality of life compared to normal hearing peers.[8]

For children with unilateral aural atresia resulting in conductive hearing losses, however, literature is unclear. These children differ from children with unilateral severe to profound SNHL, as they still hear their own voice in the affected ear, the absolute hearing loss is mostly less profound and bone conduction devices can overcome the conductive hearing deficit. Only a few studies have investigated educational performance in children with aural atresia.[9–11] They reported a higher need for individualised education plans and speech therapy and requiring additional school intervention.[9–11] Evidence is lacking on hearing-related quality of life or the language, educational or social-emotional development of these children. A lower quality of life is correlated with lower educational performance.[12] As children with aural atresia have a lower quality of life,[13] it is implied that these children show lower educational performance. Therefore, the direct relation between aural atresia and educational performance needs to be assessed. Better knowledge and understanding of the situation may help in the ongoing debate whether the use of hearing amplification or attention for guidance of children with unilateral congenital aural atresia is necessary. This could enable them to overcome the challenges that accompany congenital aural atresia.

2. Problem, Aim, Research Question

Research is still lacking on the development of children and young adults with unilateral conductive hearing loss due to aural atresia. This study aims to investigate the hearing-related quality of life of children with unilateral aural atresia, in order to be able to provide parents with a clearer image of what could be expected of their children. Additionally, general quality of life, speech and language development, educational performance, and social-emotional development will be investigated. This leads to the following research question:

- What are the hearing-related quality of life, developmental outcomes and performance of children with unilateral conductive hearing loss due to congenital aural atresia?

3. Method

Ethical consideration

The ethics committee of the University Medical Centre Utrecht (UMCU) declared that no formal approval of the detailed protocol was needed according to the Dutch Medical Research Involving Human Subjects Act (No.14-850/C).

Study design

This study had a cross-sectional cohort design in which children and young adults with unilateral conductive hearing loss due to congenital aural atresia were included. The study had a total duration of three months and took place at the department of Ear, Nose and Throat and Audiology of the Wilhelmina Children's Hospital Utrecht.

Study population

Participants were children and young adults with unilateral conductive hearing loss due to aural atresia, aged between six and 20 years old. Patients were recruited from the patient database of the Department of Plastic Surgery of the Wilhelmina Children's Hospital Utrecht after historical consultation for reasons of microtia.

In order to participate in this study, a subject was required to meet all of the following criteria: A participant must 1) be aged between six and 20 years old at the time of the study, 2) have Dutch as primary language, either themselves or their caregivers, 3) have single-sided congenital aural atresia (codes Q16.0 to Q16.4 in the International Classification of Diseases and Related Health Problems)), 4) have a hearing loss of minimal 40 dB nHL with perceptive hearing level maximum of 20 dB nHL (1000-4000 Hz) on ipsilateral side (atretic ear), measured at their last appointment with an audiological centre, 5) have a hearing level of 20 dB nHL maximum (1000-4000 Hz) on contralateral side (best ear), and 6) have given informed consent, either themselves or their caregivers. Children with syndromes that met the inclusion criteria were also included in this study. As such, there were no criteria that led to exclusion of a participant.

Procedure

Out of the database that was used to include participants, 161 children and young adults were selected based on the eligible age criteria to participate in the study. Recruitment of participants took place from December 2017 to April 2018. Participants were contacted by the researchers by physical mail, in which participants were informed and asked for consent. Participants (and their parents) were included when they met the inclusion criteria. Thereafter, they received all applicable questionnaires by physical mail. After

three weeks without returning the questionnaires, participants were contacted by phone and reminded of the study. Participants were marked as a drop-out after 6 weeks without response and analysed to investigate the reason for dropping out. Results of the questionnaires were analysed after all participants either handed in the questionnaires or have dropped out.

Variables and data measurement

Demographic data was collected by questioning age, sex, level of education of legal custodians (divided into professional education (mbo), university of applied sciences (hbo) and university (wo)), multilingualism and medical conditions or syndromes. Participants were divided into age groups, with age group 1 (younger) consisting of children between 6;0 and 10;11, age group 2 (middle) consisting of children between 11;0 and 15;11, and age group 3 (older) consisting of children and young adults between 16;0 and 20;11.

The primary outcome of this study was hearing-related quality of life measured by the Speech Spatial and Qualities of Hearing Scale (SSQ). The SSQ measures quality of hearing related activities, thereby measuring perceived hearing handicap.[14] The SSQ consists of 24 VAS-scales, scoring 0 (not at all) to 10 (perfect), covering Speech, Spatial and Quality of Hearing. The means of the VAS-scales of the three subdomains (Speech, Spatial and Quality of Hearing) are then compared to the norms.[14] For children of 6 up to 15 years a parental version is used (adapted by Karyn Galvin, and translated to Dutch by Liesbeth Royackers Labo Exp ORL, Leuven). Participants of 16 years old and above filled in the SSQ themselves.[14]

The secondary outcomes of this study were 1) general quality of life, 2) language development, 3) educational performance, and 4) social-emotional development. General quality of life was measured by the Kidscreen-27. This questionnaires assesses health-related (general) quality of life,[15] and includes five subcategories (physical well-being, psychological well-being, autonomy & parent relation, peers & social support and school environment), covered by 27 questions with a 5-point rating scale (ranging from not at all/never to extremely/always). Scores on the Kidscreen-27 are calculated by summing up the scores of the subscales, converting them to a Rasch score and converting those Rasch scores to a T-score. The scores on the questions are added per subcategory and compared to the norm scores. Participants of 11 years and older filled in the questionnaire themselves. For children younger than 11 years old, the Kidscreen-27 was filled in by parents.

Language development for children between 6 and 16 years old was measured with the Dutch version of the Children's Communication Checklist (CCC-2-NL). It measures aspects of language structure, such as vocabulary and storytelling.[16] The CCC-2-NL is divided into ten categories: speech, syntax, semantics, coherence, initiation, stereotypical language, use of context, non-verbal communication, social relations and interests. In addition, the CCC-2-NL provides three communication scales for general communication, social interaction and pragmatics. The CCC-2-NL is a parental questionnaire. In total, the CCC-2-NL consists of 70 4-point frequency scales, ranging from never to always. The scores on the items are added together per subscale and are then converted to a standard score. Their totals are compared to the norm scores of the CCC-2-NL[16], for which higher scores mean lower ability on the respective subscale. This questionnaire has been validated for Dutch children between 4 and 15;6 years old. Participants of 16 years of age and older were not evaluated on speech and language development by the lack of a validated questionnaire to assess this outcome. Age group 3 is therefore not analysed with the CCC-2-NL.

To investigate the educational performance of the participants, the participants were asked for the occurrence of grade retention, as well as the need for special education, speech therapy, special measures in class, educational assistance and hearing amplification during lifetime.

Social-emotional development was measured using the Children's Behaviour Checklist (CBCL) for participants between 6 and 18 years old.[17] The CBCL is a validated parental questionnaire that enables researchers to identify and quantify social skills and possible behavioural problems.[17,18] It consists of 113 statements with 3-point rating scales (ranging from not true to often true), where higher scores denote higher risk of behavioural problems. The items on the CBCL correspond with several subdomains: withdrawn, somatic complaints, anxious/depressed, social problems, attention problems, delinquent behaviour and aggressive behaviour. The total scores of these subdomains were compared to their norm scores. Scores on the CBCL and YSR/ASR are calculated with a scoring programme, ADM version 9.1. For participants between 6 and 18 years old, the CBCL is filled in by parents. Additionally, participants between the age of 11 and 18 years old filled in the Youth Self-Report (YSR), which is a self-report version of the CBCL. 18 to 20-year-old participants filled in the Adult Self Report (ASR), which is an adult version of the CBCL.

Data analysis

All statistical analyses were conducted using the Statistical Package for the Social Sciences (SPSS) Statistics version 24.0. Descriptive statistics were used to describe characteristics of participants. The SSQ collects quantitative numerical data on hearing-related quality of life of the participants. As such, differences between the means of subscales and age groups were calculated using a One-Way ANOVA. In case of a significant difference, an independent t-test was used to identify the differing groups.

Quantitative data of the secondary outcomes of the Kidscreen-27, CCC-2-NL and the CBCL/YSR/ASR questionnaires were compared to their norm scores. Differences between age groups were calculated using a Kruskal-Wallis test. When a Kruskal-Wallis test returned a significant difference, a Mann-Whitney U test was used to identify which groups differed from each other. As mean scores on two different subscales are paired samples, differences between subscales were calculated with a Wilcoxon signed rank test (with not-normal distribution) or a paired t-test (with normal distribution). Using a mixed models growth analysis on the age groups, an image could be construed of the development of the performance scores on the questionnaires.

Educational performance was investigated by asking for quantitative data regarding educational history and special educational needs of the participants. This data was collected and summarised. Data regarding the use of speech and language therapy and the use of hearing aids are descriptive.

4. Results

Participants

Figure 1 shows the flow chart of the inclusion procedure. Out of the 161 participants invited to participate, 37 returned informed consent and were included. In total, 20 participants returned the questionnaires, of which 19 were analysed. Their characteristics are summarised in table 1, as well as the results of the custom questionnaire for school performance. One participant did not fill in parts of the custom questionnaire.

Of the 19 participants, 10 were male and nine were female. Age ranged from 7;6 to 19;3. Age group 1 consisted of five participants, ranging from 7;6 to 10;11. Age group 2 consisted of eight participants, ranging from 11;2 to 15;9. Age group 3 consisted of six participants, ranging from 16;8 to 19;3. One participant was brought up multilingually. Eight participants have made use of hearing aids. Four children have been diagnosed with a syndrome or medical condition, namely Goldenhar syndrome (2), Attention Deficit Disorder (1) and Auriculocondylar syndrome (1). Four participants grew up in a highly educated environment, six grew up in a medium educated environment and eight grew up in a low educated environment. Five participants (26%) repeated a class. Two participants (11%) were in special education programmes. Thirteen participants (68%) have needed speech therapy. Fifteen participants (79%) have needed special measures in class, of which six (32%) needed multiple measures. Measures mentioned included orientation with their best ear towards the teacher (4), a special position in front of the class (12), educational assistance (3) and solo equipment (1).

Primary outcome: hearing related quality of life

Total scores on the SSQ subscales were calculated with means, summarised in table 2. No significant differences were found between age groups. Participants scored lower on the spatial subscale (M=5.00, sd=2.30) compared to the speech (M=6.78, sd=1.60, p= .001) and quality of hearing (M=6.98, sd=1.75, p= .001) subscales. Scores on the speech and quality of hearing subscales did not differ significantly (p= .0184).

Secondary outcomes

Mean T-scores on the Kidscreen-27 are summarised in table 3. On the physical well-being subscale, age group 3 (M=42.71, sd=7.42) scored significantly lower to age group 1 (M=62.72, sd=7.61, p=.015) and group 2 (M=55.00, sd=7.26, p=.018). On the same

subscale, age group 1 scored 1.2 standard deviations above the norm. All other mean scores fell within normal range.

The mean standard scores of the CCC-2-NL are summarised in table 4. Nearly all mean scores on the ten subscales fell within one standard deviation of the norm. This means that nearly all mean scores of the subscales fell within normal range. Mean scores for the communication scales fell within normal range as well: general communication fell between the 70th and 75th percentile, social interaction fell between the 45th and 60th percentile, and pragmatics fell between the 70th and 75th percentile. No differences were found between the age groups on any subscale of the CCC-2-NL. On the stereotypical language subscale, age group 2 scored 1.1 standard deviations above the norm.

The mean T-scores for the CBCL and YSR/ASR are summarised in table 5 and table 6, respectively. A difference was found between age groups on the attention-seeking behaviour subscale of the CBCL: the younger group (M=62.80, sd=5.40) showed higher scores than both the middle group (M=53.25, sd=4.06, p=.012) and the older group (M=55.00, sd=2.65, p=.036). Additionally, a difference was found between age groups on the withdrawn/depressed subscale of the YSR/ASR: the older group (M=60.00, sd=6.54) showed higher scores than the middle group (M=52.43, sd=2.82, p=.044). All mean scores fell within non-clinical range.

Other findings

None of the other demographic variables (Sex, level of education of caregivers, use hearing amplification, multilingualism, presence of syndromes) were found to have an effect on SSQ scores.

For the Kidscreen-27, CCC-2-NL, CBCL and YSR/ASR, some variables were found to have an effect on mean scores. Level of education of caregivers was found to have an effect on the physical well-being subscale of the Kidscreen-27 (p=.020): children who grew up in a medium educated environment (M_{medium}=55.89, sd=8.44) showed better scores compared to children from a low educated environment (M_{low}=45.71, sd=8.69) and worse scores compared to children from a highly educated environment (M_{high}=64.06, sd=6.52). In addition, level of education or caregivers was found to have an effect on the attention-seeking subscale of the YSR/ASR (p=.038): children from a low educated environment (M=56.50, sd=8.98) showed worse scores compared to children from a medium educated environment (M=51.00; sd=1.41). Use of hearing amplification had an effect on the coherence subscale of the CCC-2-NL (p=.009): children with hearing amplification (M=13.83, sd=1.47) showed worse scores than children without hearing amplification

(M_{no}=10.17, sd=2.40). Presence of syndromes had an effect on the use of context subscale of the CCC-2-NL (p=.045): children with syndromes (M=15.67, sd=2.08) showed worse scores compared to children without a syndrome (M=11.11, sd=2.52). Sex and multilingualism had no effect on any scores.

5. Discussion

This study aimed to investigate the hearing-related quality of life, developmental outcomes and performance of children and young adults with unilateral conductive hearing loss due to aural atresia.

The outcome of the hearing related quality of life in the presented study was compared to data of normal-hearing children and subjects with unilateral SNHL,[19] because of the lack of norm data out of the general population in this age. As the scores of the current study (M_{speech}=6.78, $M_{spatial}$ = 5.00, M_{QoH}=6.98) are lower than the scores of the normal-hearing children in Sangen et al. (M_{speech}=9.0, $M_{spatial}$=8.5, M_{QoH}=9.0), it is assumed that the participants of the current study scored significantly lower than the normal-hearing children in Sangen et al. Whether the scores of this study differed significantly from the scores of the children with SNHL is unknown.

In nearly all instances, mean scores for general quality of life (Kidscreen-27), language development (CCC-2-NL) and social-emotional development (CBCL, YSR/ASR) fell within normal range. This suggests that children with congenital conductive hearing loss develop comparatively to normal-hearing peers. No effect could be found for use of hearing amplification, age, sex, level of education of parents, multilingualism or syndromes. Some differences were found in outcomes between age groups. The group of 16 to 20 year scored significantly lower to children below 16 years on physical well-being as a subscale of the Kidscreen-27 test. This suggests that their physical well-being is worse compared to younger participants. Additionally, the older group of 16 years and older scored significantly higher than the younger groups (6 to 15 years) on the withdrawn/depressed subscale of the CBCL, meaning that these participants tend to exhibit withdrawn behaviour more often than their younger peers. Lastly, the middle group of 11 to 15 year scored significantly higher than the older group on the attention-seeking behaviour subscale of the YSR/ASR. This means that the middle group tends to exhibit attention-seeking behaviour more often than their older peers. However, all mean score still fell within non-clinical range. This raises questions about the validity and relevance of these findings. It is possible that any actual differences do not present themselves due to the wide range of the norms and the limited size of the study population.

While the literature on the subject of unilateral congenital aural atresia is limited, some comparisons can be made. Notably, while aural atresia has been suggested to have a detrimental effect on general quality of life[13], no such effect has been found in the current

study. Likewise, the current study did not find an effect of aural atresia on language development, even though language problems have been found in children with SNHL.[5] The lack of an effect for these domains in the current study could be attributed to the low number of participants and the limited statistical power of non-parametric tests.

Regarding educational performance, two variables were of note. 26% repeated a class, of which the majority around group 3, when children first learn to read. According to the Central Planning Bureau (Centraal Planbureau)[20], around 5% of Dutch children repeat group 3. The percentage of children in this study who repeated a class in that period is therefore higher than is to be expected. This outcome is explained in children with SNHL with educational difficulties by the encountered language problems.[5–7] These problems were also expected in children with unilateral conductive hearing loss of magnitudes seen in unilateral aural atresia. Additionally, similar to children with SNHL, the use of special measures in education and the need for language therapy is prevalent in these children. This implies that, despite the differences between SNHL and unilateral conductive hearing loss due to aural atresia, both groups of children encounter similar problems regarding educational performance. The findings of the current study are in line with previous studies on the subject.[9–11]

To our knowledge this is the first study reporting on language development and social-emotional development in which validated questionnaires were used to assess outcomes. However, our study has some limitations. A small number of participants was included. This was due to the low response rate of the target population (39%). This response rate is lower than the proposed norm of 60% ± 20.[21] A low response rate could lead to lower validity, reliability and dependability of the results.[21]
Additionally, while the differences that were found were significant, all mean scores fell within normal or non-clinical range. It could be discussed whether the effects thereby are of any clinical importance. A tendency towards certain behaviour is observed at most, but due to the small number of participants any found tendencies are tentative at best. Therefore, the results of this study imply that children with unilateral conductive hearing loss due to aural atresia develop normally compared to normal-hearing peers. Still, it must be taken into consideration that the participants of this study included eight children (42%) who received hearing amplification at a prior age. While no significant differences were found in outcomes between users and non-users of hearing amplification, it could still be seen that children with hearing amplification showed higher performance scores on the questionnaires. A study with a large population could identify possible clinically relevant

differences that support the use of hearing amplification in this group as a requirement for performances compared to normal hearing peers.

Due to the small number of participants, a mixed models growth analysis of the scores on the questionnaires could not be performed. This limits the validity of this study. Still, this study included more participants than other studies for this population group. Therefore, it is still an important addition to the available knowledge on the subject of unilateral aural atresia.

6. Conclusion

Children with unilateral aural atresia are suggested to have a lower hearing-related quality of life compared to normal-hearing peers and show a higher need for educational assistance. Regarding language development and social-emotional development, however, children and young adults with aural atresia seem to develop normally. Due to the small number of participants included in this study, any significant differences that were found have to be confirmed in future studies to ascertain their veracity.

Children with unilateral conductive hearing loss due to aural atresia seem to encounter similar educational problems as children with SNHL. It is therefore prudent to provide assistance to children with aural atresia regarding educational endeavours.

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8. Figures and Tables

Figure 1: Flow diagram of inclusion procedure.

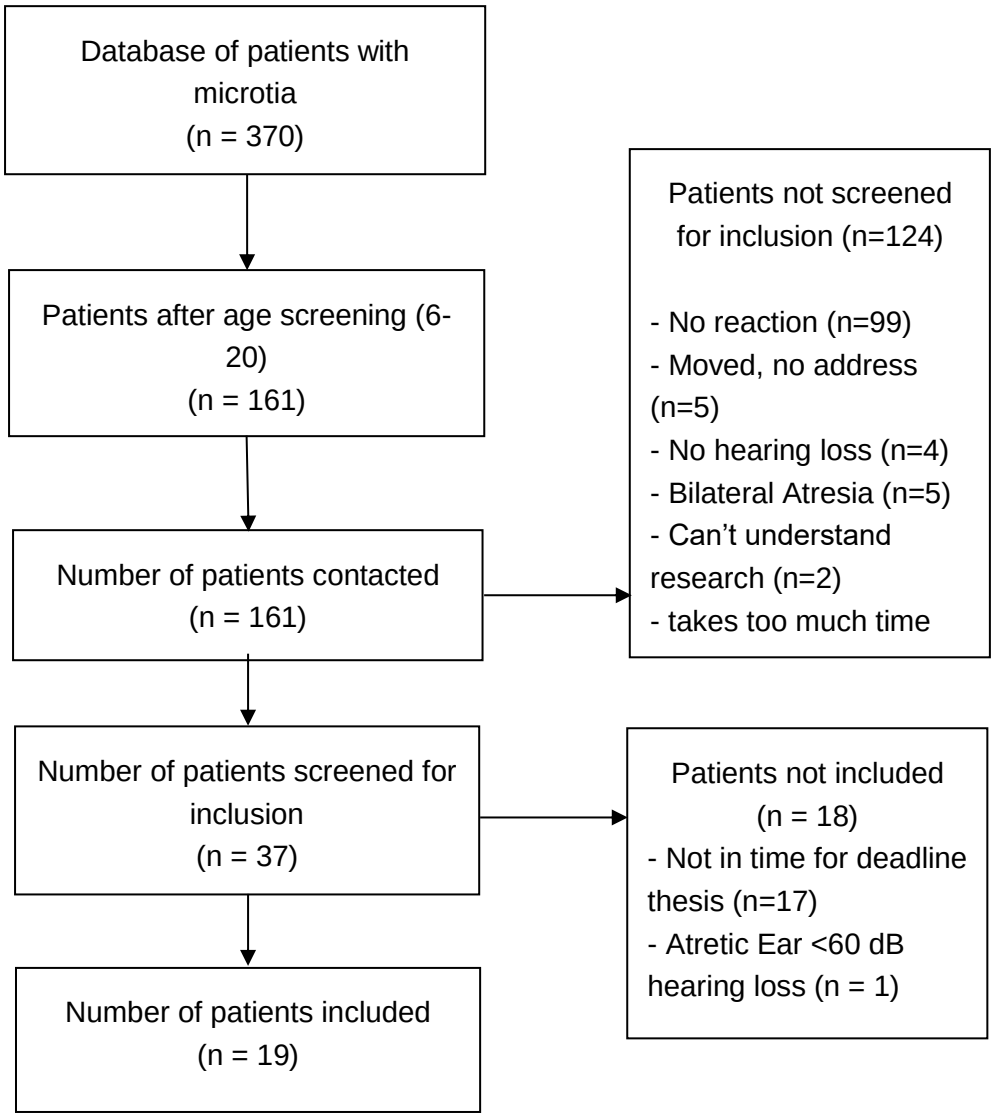

Table 1: Demographic characteristics of participants and outcomes of custom questionnaire on school performance in frequency and (percentages) (N=19)

			Total	Age group 1 (N=5)	Age group 2 (N=8)	Age group 3 (N=6)
Median age in months (iqr)			155 (70)	106 (35)	153 (49)	203.5 (29)
Sex (%)	-	Male	10 (52.6)	4 (80.0)	2 (25.0)	4 (66.7)
	-	Female	9 (47.4)	1 (20.0)	6 (75.0)	2 (33.3)
Repeating of classes (%)	-	No	14 (73.7)	5 (100.0)	5 (62.5)	4 (66.7)
	-	Yes	5 (26.3)	0 (0.0)	3 (37.5)	2 (33.3)
Use of Special Education (%)	-	No	17 (89.5)	5 (100.0)	7 (87.5)	5 (83.3)
	-	Yes	2 (10.5)	0 (0.0)	1 (12.5)	1 (16.7)
Language Therapy (%)[a]	-	No	5 (26.3)	2 (40.0)	3 (37.5)	0 (0.0)
	-	Yes	13 (68.4)	3 (60.0)	5 (62.5)	5 (100.0)
Use of hearing amplification (%)[a]	-	No	10 (55.6)	2 (40.0)	5 (62.5)	3 (60.0)
	-	Yes	8 (44.4)	3 (60.0)	3 (37.5)	2 (40.0)
Use of special measures in education (%)[a]	-	No	3 (15.8)	2 (40.0)	0 (0.0)	1 (20.0)
	-	Yes	9 (47.4)	2 (40.0)	4 (50.0)	3 (60.0)
	-	Yes, multiple	6 (31.6)	1 (20.0)	4 (50.0)	1 (20.0)
Highest level of education (%)[a]	-	Mbo	8 (44.4)	0 (0.0)	4 (50.0)	4 (80.0)
	-	Hbo	6 (33.3)	2 (40.0)	3 (37.5)	1 (20.0)
	-	Wo	4 (22.2)	3 (60.0)	1 (12.5)	0 (0.0)
Multi-lingualism (%)	-	No	18 (94.7)	4 (80.0)	8 (100.0)	6 (100.0)
	-	Yes	1 (5.3)	1 (20.0)	0 (0.0)	0 (0.0)
Presence of syndromes (%)	-	No	15 (78.9)	4 (80.0)	6 (75.0)	5 (83.3)
	-	Yes	4 (21.1)	1 (20.0)	2 (25.0)	1 (16.7)

[a] N = 18, One participant refused to fill in part of the custom questionnaire. iqr=interquartile range.

Table 2: Speech, Spatial and Quality of Hearing scale mean scores and (standard deviations) per subscale in total and by age groups (N=19)

	Total	Age group 1	Age group 2	Age group 3
Speech Subscale	6.78 (1.60)	7.28 (1.49)	6.95 (1.73)	6.14 (1.57)
Spatial Subscale	5.00 (2.30)	4.70 (2.09)	5.18 (2.14)	4.99 (3.01)
Quality of Hearing subscale	6.98 (1.75)	7.34 (2.09)	7.55 (1.48)	5.93 (1.59)

Table 3: Mean T-scores and (standard deviations) per subscale of the Kidscreen-27 in total and by age group (N=18?)

	Total	Age group 1	Age group 2	Age group 3
Physical	53.73 (10.42)	62.72 (7.61)	55.00 (7.26)	42.71 (7.42)
Psychological	50.25 (8.89)	51.60 (7.81)	53.25 (9.58)	44.12 (6.86)
Parents	53.62 (7.09)	54.16 (11.83)	52.00 (5.38)	55.67 (3.49)
Peers	52.01 (10.00)	50.36 (12.35)	56.69 (9.57)	47.13 (6.22)
School	54.50 (7.54)	54.69 (11.96)	56.49 (6.38)	51.14 (2.23)

Mean norm score is 50 (sd=10).

Table 4: Mean standard scores on the CCC-2-NL items in total and by age group (N=12)

	Total	Age group 1	Age group 2
Speech[a]	10.75 (4.09)	10.20 (3.49)	11.14 (4.71)
Syntax[a]	10.42 (3.03)	10.40 (3.58)	10.43 (2.88)
Semantics[a]	11.17 (2.52)	11.60 (2.70)	10.86 (2.55)
Coherence[a]	12.00 (2.70)	12.60 (2.07)	11.57 (3.16)
Initiation[a]	9.92 (3.09)	10.00 (3.24)	9.86 (3.24)
Stereotypical language[a]	12.67 (2.84)	11.60 (3.13)	13.43 (2.57)
Use of context[a]	12.25 (3.11)	12.60 (1.82)	12.00 (3.92)
Non-verbal[a]	12.08 (2.81)	12.00 (4.06)	12.14 (1.86)
Social relations[a]	11.33 (2.54)	12.20 (3.03)	10.71 (2.14)
Interests[a]	10.83 (2.48)	11.40 (2.19)	10.43 (2.76)
General communication[b]	91.25 (18.28)	91.00 (20.10)	91.43 (18.52)
Social Interaction[c]	0.33 (10.50)	2.00 (7.11)	-0.86 (12.83)
Pragmatics[d]	46.92 (8.76)	46.20 (10.85)	47.43 (7.85)

[a]mean norm is 10 (sd = 3). [b]range 44-122, 80 corresponds to 50th percentile. [c]range -19 to 19, 0 corresponds to 50th percentile. [d]range 19-63, 40 corresponds to 50th percentile.

Table 5: Mean T-scores and (standard deviations) on the CBCL in total and by age group (N=16)

	Total	Age group 1	Age group 2	Age group 3
Anxiety	53.81 (5.31)	55.80 (7.33)	54.00 (4.54)	50.00 (0.00)
Withdrawn/Depressed	55.44 (5.59)	53.60 (4.98)	54.88 (5.77)	60.00 (5.20)
Somatic	58.38 (7.62)	58.20 (10.16)	57.88 (7.00)	60.00 (7.21)
Social	54.56 (5.62)	59.00 (7.14)	52.88 (4.05)	51.67 (2.08)
Thoughts	57.75 (7.18)	59.00 (5.48)	58.00 (8.45)	55.00 (7.81)
Attention seeking	56.56 (5.98)	62.80 (5.40)	53.25 (4.06)	55.00 (2.65)
Delinquent behaviour	52.81 (3.27)	55.00 (4.30)	51.38 (1.69)	53.00 (3.61)
Aggressive	53.88 (5.24)	56.80 (7.16)	52.38 (3.89)	53.00 (4.36)

Mean norm is 50 (sd=10). A score above 68 denotes clinical range.

Table 6: Mean T-scores and (standard deviations) on the YSR/ASR in total and by age group (N=13)

	Total	Age group 2	Age group 3
Anxiety	53.85 (4.47)	53.86 (5.18)	53.83 (3.97)
Withdrawn/Depressed	55.92 (6.10)	52.43 (2.82)	60.00 (6.54)
Somatic	57.38 (10.51)	58.86 (14.44)	55.67 (2.88)
Social	54.23 (5.33)	52.00 (2.16)	56.83 (6.88)
Thoughts	55.46 (6.12)	56.14 (5.93)	54.67 (6.80)
Attention seeking	54.46 (7.40)	55.43 (10.10)	53.33 (2.50)
Delinquent behaviour	51.92 (2.14)	51.57 (2.82)	52.33 (1.03)
Aggressive	51.85 (3.85)	51.86 (4.91)	51.83 (2.56)

Mean norm is 50 (sd=10). A score above 68 denotes clinical range.