Original Investigation

Effects of Aural Atresia on Speech Development and Learning
Retrospective Analysis From a Multidisciplinary Craniofacial Clinic

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IMPORTANCE Aural atresia (AA) is associated with maximal conductive hearing loss in affected ears, and children with bilateral AA require amplification. Some recent research has suggested an increased risk for speech and learning problems among children with unilateral sensorineural hearing loss.

OBJECTIVE To investigate whether increased risk for speech and learning problems exists among children with AA.

DESIGN Retrospective medical record review.

SETTING Multidisciplinary craniofacial clinic.

PARTICIPANTS Children with unilateral or bilateral AA.

INTERVENTIONS Records review, including evaluations by audiologists, speech pathologists, and psychologists.

MAIN OUTCOME MEASURES Rates of speech and/or language delay, prevalence of speech therapy and educational interventions, and parental report of psychosocial problems.

RESULTS A total of 74 patients were identified who met inclusion and exclusion criteria: 48 with right-sided AA, 19 with left-sided AA, and 7 with bilateral AA. Children with AA demonstrated high rates of speech therapy (86% among bilateral, 43% among unilateral). Reports of school problems were more common among children with right-sided AA (31%) than those with left-sided AA (11%) or bilateral AA (0%) (P = .06). Educational interventions were common in all groups (33% right, 21% left, 43% bilateral). In the case of bilateral AA, all children who received additional interventions were enrolled in schools for the hearing impaired, without any identified learning deficiencies.

CONCLUSIONS AND RELEVANCE Children with unilateral AA may be at greater risk of speech and learning difficulties than previously appreciated, similar to children with unilateral sensorineural hearing loss. Whether amplification may alleviate this risk is unclear and warrants further study.

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Aural atresia (AA) is a congenital absence or stenosis of the external auditory canal with variable middle ear anomalies. It is almost always accompanied by a malformed (microtia) or absent (anotia) external ear.[1] Atresia and microtia are common congenital malformations, with the incidence of microtia reported to be 0.5 to 3 per 10 000 live births, and AA reported in 55% to 93% among individuals with microtia.[1-3] Microtia is unilateral in most patients, with a right-sided and male predominance.[1] Children with bilateral hearing loss are known to be at increased risk of speech and language delays as well as poor performance in school.[4,5] Children with bilateral AA typically demonstrate bilateral maximal conductive (ie, moderately severe) hearing loss and are generally assumed to have similar speech-language and school problems, though this has not been specifically studied. In contrast, the clinical significance of unilateral hearing loss (UHL) is not universally recognized.[6] Traditionally, children with UHL were thought to develop normally without any major sequelae, and most have not received amplification. Several small studies of children with UHL have shown delays in word acquisition in younger children, increased grade failure rates, and a higher prevalence of special educational interventions; however, these results have been inconsistently demonstrated.[7-14] Some studies have demonstrated poor hearing aid compliance among children with UHL, though others have shown greater than 80% acceptance.[12,15] Lieu et al[16] conducted a prospective case-control study comparing scores on the Oral and Written Language Scales (OWLS) test in children with UHL with normal-hearing siblings. This study demonstrated significant deficits among children with UHL in language comprehension, oral expression, and oral composite score. These children were also more likely to have received speech or language therapy and to have an individual education plan (IEP) at school. Longitudinally, a subgroup of these children with UHL were found to improve their language scores, whereas their academic and behavioral problems persisted.[17] While these findings are significant, it is unclear if they can be extrapolated to children with unilateral AA because most of the children in those studies had profound sensorineural loss (SNHL). It is also unclear whether the side of the AA might impact development of speech and language.

The purpose of the present study was to use assessments by speech therapists and psychologists during visits to a craniofacial clinic to investigate the potential impact of AA on school performance and successful acquisition of speech and language. We hypothesized that children with AA would demonstrate delays in language acquisition and require speech therapy at a rate greater than that of the general population and that these delays would adversely affect school performance, as reported by their parents. We further hypothesized that children with unilateral AA would experience fewer problems than those with bilateral AA but more than the general population. Finally, one of the goals of any multidisciplinary craniofacial clinic is to track the developmental progress of its patients. Little is known, however, about the actual attendance patterns of children with AA at craniofacial clinics. We collected data to reflect the duration and frequency of clinic visits and the age range of the patients included in the study.

Methods

A waiver of informed consent was obtained from the institutional review board of Washington University in St Louis, Missouri. We performed a retrospective medical chart review of patients with AA, both unilateral and bilateral, treated in the pediatric craniofacial multidisciplinary clinic at the St Louis Children's Hospital between 1975 and 2010. We attempted to collect data for children with microtia without AA (ie, normal hearing) as a control group, but there were too few. Data were collected from the evaluations performed by audiologists, speech pathologists, and psychologists.

Inclusion Criteria

We searched the craniofacial clinic patient database using search terms *aural atresia* and *microtia*. Children aged 2 to 12 years carrying a diagnosis of unilateral or bilateral AA were included.

Exclusion Criteria

Subjects with any of the following characteristics were excluded from the study:

1. Younger than 2 years or older than 12 years at the time of the clinic visit (except for purposes of calculations regarding clinic attendance);
2. Lacked audiogram or speech pathologist examination data;
3. Carried other potentially confounding diagnoses, including developmental delay, cleft lip or palate, or other syndrome, with the exception of hemifacial microsomia;
4. Had 40-dB or less hearing level (HL) in an atretic ear, or 20-dB or greater HL in unaffected ears in children with unilateral AA; and/or
5. History of recurrent or chronic otitis media requiring tympanostomy tube placement in the unaffected ear.

Outcome Variables

Demographic variables included age and sex. To understand clinic usage patterns for these patients, we collected data reflecting patient age at first and last visits to the clinic as well as total number of visits.

Audiogram values included thresholds in dB HL for both air and bone conduction and pure tone average. Amplification data included type of amplification and age at initiation. Otologic history included history of recurrent or chronic otitis media and prior atresiaplasty.

Data collected from speech pathologist evaluations included presence of articulation or language errors, voice or resonance abnormalities, and history of speech therapy with duration and age at initiation.

Parental report of school performance was reviewed, especially problems with learning, discipline, or attention. Utilization of additional educational resources was recorded, including brief trial interventions to see if the patient's performance would catch up (*response to intervention*), formal structured instruction (*tutoring*, *individual education plan*), or enrollment in special education outside the mainstream classroom. Psychologic data included history of any psychiatric diagnosis or treatment.

Table 1. Demographic and Audiologic Profile of Patients with AA

Characteristic	Right AA (n = 48)	Left AA (n = 19)	Bilateral AA (n = 7)	Total (n = 74)
Sex, No. (%)				
Male	30 (63)	9 (47)	2 (29)	41 (55)
Female	18 (38)	10 (52)	5 (71)	33 (45)
Race/ethnicity, No. (%)				
African American	3 (6)	1 (5)	1 (14)	5 (7)
White	38 (79)	15 (79)	3 (43)	56 (76)
Asian	2 (4)	1 (5)	1 (14)	4 (5)
Latino	2 (4)	0	0	2 (3)
Unknown	3 (6)	2 (11)	2 (29)	7 (9)
Clinic visits, mean (SD)				
Total	3.1 (1.7)	2.8 (1.3)	2.3 (1.7)	2.9 (1.6)
Age at last visit	9 (3.1)	7.8 (3.4)	7.1 (3.1)	8.5 (3.2)
Pure tone average, mean (SD)[a]	(n = 44)	(n = 18)	(n = 5)	(n = 67)
Affected ears	64 (8)	64 (8)	60 (13)	63 (8)
Unaffected ears	6 (6)	9 (4)	NA	7 (6)

Abbreviations: AA, aural atresia; NA, not applicable; PTA, pure tone average.

[a] If insufficient data for PTA, then the average of 2 frequencies or speech reception threshold were used, if available; otherwise the patient was excluded. While 7 patients were excluded from PTA calculations for affected ears, all patients had adequate data for PTA of unaffected ears. No demographic or audiologic comparisons reached statistical significance.

Statistical Analysis

Descriptive statistics were calculated, including averages, frequencies, proportions, and standard deviations. Statistical analyses were performed using χ^2 analysis, with statistical significance considered to be $\alpha = 0.05$. Subjects were grouped according to AA status (right, left, or bilateral) for comparison.

Results

From an initial pool of 168 subjects, 74 were included in the analysis: 48 with right AA, 19 with left AA, and 7 with bilateral AA. Most of the patients excluded from the study had inadequate medical records (n = 90), having been added to the clinic database on referral but never attending. Three children had microtia without AA, and thus did not have associated hearing loss; however, they were too few in number to serve as a control group. No other cohort of children attending the craniofacial clinic was deemed an appropriate control because none was without speech and/or learning issues (eg, cleft palate, craniosynostosis)[18] One patient was excluded because of a submucous cleft contributing to markedly hypernasal speech.

Demographic data are listed in **Table 1**. No statistical differences were identified between groups in any demographic category. There was a slight overall male preponderance, and over twice as many cases of right AA as left AA, both of which agree with findings of prior studies on AA.[3,19] There were no differences between groups in attendance patterns: subjects attended the clinic an average of 2 to 3 times and generally stopped attending at around ages 7 to 9 years.

Nearly all subjects demonstrated pure tone averages around 60 dB HL in the affected ear, consistent with maximal conductive loss, and normal hearing (pure tone averages less than 20 dB HL) in unaffected ears. Bone lines for atretic ears consistently demonstrated purely conductive hearing loss in almost all patients, though these data were missing in a total of 5 patients (4 right AA, 1 left AA). Additionally, 4 patients demonstrated evidence of a sensorineural component to their hearing loss (2 right AA, 1 left AA, 1 bilateral AA), all of which was mild, with the exception of that of the patient with bilateral AA, who had mild to moderately severe SNHL in 1 ear. Exclusion of these patients did not substantively change statistics in any category, and they were included so as to reflect the overall AA population.

Amplification was universal in subjects with bilateral AA (7 of 7; mean age, 1.5 years), while few subjects with right AA (3 of 48) and none with left AA received amplification. When aggregated, only 3 of 67 subjects with unilateral AA received amplification (4%), at an average age of 5.5 years. Of these, 1 received an FM system (frequency modulated) and 2 received softband Bahas (Cochlear Ltd). All 3 children with unilateral AA received amplification during or after 2004. One child with a softband Baha went on to receive a permanent abutment, while the other abandoned the Baha. All but 1 of the children with bilateral AA received a bone conduction aid by age 2 years, and several underwent atresiaplasty. Details of those with amplification are listed in **Table 2**. The number of children who received amplification and/or underwent atresiaplasty was insufficient for a statistical comparison with those who did not.

A high percentage of subjects in every group had received speech therapy (**Table 3**). While the rates in the right AA and left AA groups were comparable at roughly 40%, nearly every child in the bilateral AA group had received speech/language therapy. Indeed, the only child in the bilateral AA group who had not received speech/language therapy was 2 years old at the time of her only clinic visit. Prevalence of articulation errors and language errors identified by speech pathologists during clinic visits roughly correlated with prevalence of speech/language therapy, and the differences between groups were significant (Table 3). Direct comparisons between right AA and left AA groups were not statistically significant.

Information regarding school performance was obtained by reviewing clinic notes of parental interviews performed by psychologists and clinic nurses. Thirty-one percent of children with

right AA experienced learning difficulties in school (n = 15), compared with 11% with left AA (n = 2) and none with bilateral AA (**Table 4**). Overall comparisons as well as direct right AA to left AA comparisons did not reach statistical significance. Problems with discipline and attention were uncommon in all groups. Thirty-one percent of children required additional school intervention (n = 22), and rates were similar between groups. Of note, the 3 children with bilateral AA who were coded as having received school intervention were thus coded because they were enrolled in a school for the deaf and not for any interventions initiated based on performance issues.

Review of psychiatric histories by clinic psychologists showed that 3 of 48 children with right AA had been diagnosed as having attention deficit hyperactivity disorder, and 1 child with bilateral AA had received counseling for selective mutism in school.

Table 2. Individual Amplification Profiles

Patient Age at Amplification, y[a]	Year[b]	Type of Amplification
Right AA		
2	2008	Baha (Cochlear Ltd) (softband, stopped after <1 y)
10	2008	Baha (softband, then abutment)
5	2004	FM system
Bilateral AA		
0.5	1993	Bone conduction aid, later transitioned to bilateral air conduction (no mention of atresiaplasty)
2	1975	Bone conduction aid, then air conduction aid after atresiaplasty age 6 years
5	1996	FM system, atresiaplasty age 5 years, no mention of any amplification prior to atresiaplasty
0.5	1984	Bone conduction aid, then right air conduction aid after right atresiaplasty at age 4 years
1	1995	Bone conduction aid (no follow-up)
0.5	1999	Bone conduction, then Baha
2	2002	Bone conduction (brief follow-up)

Abbreviations: AA, aural atresia; FM, frequency modulated.

[a] Age at amplification was recorded in years, rounded to nearest 0.5 year.

[b] Year indicates amplification initiated.

Discussion

While this study is preliminary, there were several intriguing findings that bear further investigation. First, we found that approximately 40% children with UHL required speech/language therapy (n = 29). This finding mirrors the findings of prior research: Lieu et al[16] found that 42% of children with UHL had received speech therapy, compared with 22% of sibling controls with normal hearing, a strongly significant difference. Taken together, these findings suggest that children with UHL, regardless of cause, are at increased risk of delays in speech/language development warranting intervention.

Second, these findings suggest that children with right AA may experience greater problems with speech development than children with left AA. While the magnitude of the difference in speech therapy rates was not statistically significant, the children with right AA trended toward younger age at initiation of speech therapy and higher rates of articulation and

Table 3. Speech Therapy Among Study Patients

Characteristic	Right AA (n = 48)	Left AA (n = 19)	Bilateral AA (n = 7)	Total (n = 74)	*P* Value[a]
Prevalence, No. (%)[b]	22 (46)	7 (37)	6 (86)	35 (47)	.08
Age at initiation, mean, y[c]	4.3	5.6	2.8	4.3	.19
In-clinic evaluation, No. (%)[d]					
Articulation errors	17 (35)	4 (21)	6 (86)	31 (42)	.01
Language errors	15 (31)	3 (16)	5 (71)	23 (31)	.03

Abbreviation: AA, aural atresia.

[a] *P* values obtained using χ^2 test, with all subgroups included. Direct comparisons of right and left atresia groups were not statistically significant.

[b] Prevalence of parental report of prior or current speech therapy

[c] Age at initiation calculation based on available data (18 of 22 right AA, 5 of 7 left AA, 3 of 6 bilateral AA).

[d] In-clinic evaluation refers to the frequency of pathologic articulation errors or language errors detected on speech sample in clinic by speech pathologist.

Table 4. Parental Report of Problems in School

	Study Patients, No. (%)				*P* Value	
Characteristic	Right AA (n = 48)	Left AA (n = 19)	Bilateral AA (n = 7)	Total (n = 74)	All Groups	Right vs Left
Learning	15 (31)	2 (11)	0	17 (23)	.06	.08
Discipline	7 (14)	0	1 (14)	8 (11)	.21	.08
Attention	7 (14)	2 (11)	0	9 (12)	.53	.66
Received school intervention[a]	16 (33)	4 (21)	3 (43)[b]	23 (31)	.50	.30

Abbreviation: AA, aural atresia.

[a] School intervention denotes history of in-school tutoring, attendance in "resource room," enrollment in full-time special education, or other in-school intervention intended to improve the student's performance.

[b] The 3 children with bilateral AA who were coded as having received school intervention were thus coded because they were enrolled in a school for the deaf and not for any interventions initiated based on performance issues.

language errors when evaluated by a speech therapist, as summarized in Table 4. Although none of these findings achieved statistical significance, they all align in the same direction and include both historical data (history of speech therapy) and speech pathologist evaluations at the time of clinic visits.

The overall rate of parental report of school performance problems for children with any AA was 31% (n = 23) and 30% (n = 20) for children with unilateral AA (Table 4). No child with bilateral AA was reported as having problems with school performance, and we theorize that this may be because their impairment was more fully appreciated and addressed, and their learning environment sufficiently modified to allow them to flourish; others have speculated the same.[20] Interestingly, children with right AA trended toward more problems with learning in school than did children with left AA (31% [n = 11] vs 11% [n = 2]) (P = .08), similar to prior research on the impact of UHL. Oyler et al[13] reported in 1988 a 23.5% grade failure rate among children with UHL in a large local school district, compared with a 2% rate for the general population. Additionally, nearly 35% of children with right-sided UHL had repeated a grade, compared with 6.7% of children with left-sided UHL. Similarly, Hartvig-Jensen et al[21,22] reported significantly poorer performance in children with right-sided UHL on verbal subtests in a battery of psychological tests compared with children with left-sided UHL, as well as poorer performance on interrupted speech tests in background noise.

Prior research in the fields of neuroscience and audiology provide some possible explanations for these findings. Binaural hearing has been clearly shown to be superior to unilateral hearing for comprehension, driven by the phenomena of binaural summation, binaural squelch, and the head shadow effect.[23,24] When sound is perceived in both ears simultaneously, it is perceived as louder by the brain than if it were conveyed at the same intensity to just 1 ear, an effect known as *binaural summation*. While binaural summation implies an increase in perceived gain, *binaural squelch* describes the brain's ability to filter out unimportant background noise to focus on the sounds of interest, thus improving the clarity of sound. Binaural summation and binaural squelch are products of the brain's ability to further analyze sounds when perceived from both ears at once, and this process is lost when hearing occurs in only 1 ear. Furthermore, for persons with UHL, sounds emanating from a point lateral to the bad ear will be shielded from the good ear by the head, a phenomenon known as the *head shadow effect*. Thus, despite having 1 normally functioning ear, a person may experience significant functional hearing loss from a loss of higher order processing in the brain, a problem further compounded if the sound is adversely located. Similarly, our finding of potentially worse speech and learning results for children with right AA is in line with prior work on sidedness in auditory processing. The phenomenon of *right ear advantage* was first described over 50 years ago[25] and has been demonstrated in multiple settings, including in cochlear implantation.[26] The right ear advantage describes better performance on a variety of mental tasks (comprehension, memory, execution, etc) when instructions are received in the right ear as opposed to the left. Neuroscientists have attributed the right ear advantage to contralateral cortical routing of acoustic signal, with the left auditory cortex being dominant in language processing. The concepts of binaural summation, binaural squelch, and right ear advantage appear at least partly to explain the deficits observed in those with UHL and the relatively worse outcomes for children with right-sided loss, though further investigation is warranted.

If children with unilateral AA have speech and learning consequences from their hearing loss, the question remains whether any interventions may correct or prevent these effects. Atresiaplasty is a time-honored procedure for correction of AA-related conductive hearing loss. Gray et al[27] demonstrated that patients undergoing AA repair show improvement in hearing in noise in a variety of signal/noise configurations, suggesting that patients with AA are able to experience binaural hearing once the atretic ear is rehabilitated, though there was a negative association with age. Breier et al[28] demonstrated an advantage in the nonatretic ear among patients with unilateral AA that persisted after atresiaplasty, even correcting for mild persistent hearing loss in the atretic ear. Younger age at operation correlated strongly with improved symmetry, particularly when atresiaplasty occurred before the age of 12 years. Interestingly, the 2 patients with bilateral AA included in the study showed a small advantage to the ear repaired first (left). These findings agree with older studies that demonstrated both objective and subjective hearing and quality-of-life benefits of atresiaplasty in patients with unilateral AA.[6,29] Atresiaplasty is a technically difficult and operator-dependent surgery, however, and is limited by the need to wait until after microtia repair, generally undertaken after age 5 years. Bone conduction aids were often used in the past, but are bulky and therefore socially undesirable, and are consequently less commonly used, particularly in patients with UHL. The advent of the Baha bone conduction implant added another option for patients with conductive hearing loss. Multiple studies have shown excellent results in speech recognition and improved hearing in noise in patients with conductive hearing loss, though sound localization results have been mixed.[30-32] Studies investigating outcomes with the Baha in conductive UHL have all been quite small, and further research is needed.

This preliminary study has several limitations stemming from its retrospective design. Selection bias occurs when children with AA who experience developmental difficulties are more likely to attend a craniofacial clinic and more likely to return multiple times. The data were not initially collected for the purpose of this study, so the assessment and recording of patients' speech and learning progress, while it followed standard practices, was not standardized for the study. Similarly, parental assessment or recollection (eg, history of speech therapy) may be prone to inaccuracy. Finally, the study is underpowered because of the small numbers available.

While acknowledging its limitations, we believe that this study contains several intriguing hints that may guide future work. Children with unilateral AA appear to have significant risk for speech and language delays requiring speech therapy, consistent with prior work focused on UHL of any cause. Children with right AA may be at further risk of poor performance in school owing to loss of the right ear

advantage and lack of early amplification and educational interventions that children with bilateral AA usually receive. There has been little investigation of the role of amplification in children with UHL, and future prospective studies should investigate what benefits these children may derive from early amplification. Finally, in our experience, children with AA tended to attend craniofacial clinic 2 to 3 times and then stopped attending at around age 7 to 9 years, likely because their aural reconstructions were complete or they had elected not to undergo reconstruction. In light of these findings, we would advocate close monitoring of children with AA for signs of speech or academic struggles and also advocate a low threshold for intervention, including amplification for children with unilateral AA. Craniofacial clinics should also increase efforts at retention and at achieving more long-term follow-up among these patients to facilitate identification of and intervention for any developmental difficulties that these children may encounter.

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