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Review Article

A meta-analysis of the long-term hearing outcomes and complications associated with atresiaplasty

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ABSTRACT

Objective: To summarize peer-reviewed literature to evaluate the stability of long-term hearing outcomes with prolonged follow-up, and describe the incidence of complications related to atresiaplasty.
Design: A literature search of EMBASE, MEDLINE, PubMed, Google Scholar, and Web of Science was performed to identify studies of congenital aural atresia (CAA).
Study sample: Selected studies were published between 1997 and 2014. The 19 studies covered 964 CAA ears.
Results: Stenosis and bony regrowth occurred in 14.2% (range from 0% to 30.3%). Lateralization of tympanic membrane (TM) occurred in 7.5% (range from 0% to 18.2%). Facial nerve palsy occurred in 0.5% (range from 0% to 5.0%). Six studies used Air-bone Gap (ABG) to evaluate the short-term and long-term hearing outcomes. The hearing outcomes were performed using meta-analysis, there was no significantly heterogeneity (I^2 = 0%, p = 0.997), and there was a degradation of hearing outcomes with prolonged follow-up (RR = 1.13, 95%CI: 0.98–1.30).
Conclusion: Nearly all studies focus on postoperative stenosis and bony regrowth, but some studies neglect lateralization of TM. Based on available data, which shows a lack of major complications, such as facial nerve palsy, atresiaplasty is a safe procedure. Our meta-analysis indicated that there was a degradation of hearing outcomes with prolonged follow-up.

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1. Introduction

The term CAA is used to describe a spectrum of otologic malformations. Atresia anatomically implies an isolated, narrow or stenotic canal and is often associated with various middle ear and auricular deformities [1]. CAA results from abnormal embryologic development of the first branchial arch, with an incidence of one in 10,000 to one in 20,000 births [2]. The traditional treatment is atresiaplasty, to create a patent external aural canal (EAC) and meatus and to provide hearing ability [3].

Not all patients with aural atresia are candidates for atresiaplasty. The anatomic variability, such as ectopic facial nerve or absence of the stapes, in aural atresia makes surgical correction more challenging [4]. The Jahrsdoerfer grading scale, proposed in 1992, assigns an anatomical score for the atretic ear based on the presence or absence of 9 structures. The higher the Jahrsdoerfer grading scale score, the better the chance for normal or near-normal postoperative hearing outcomes [5]. In fact, only approximately 50% of patients with CAA are currently candidates for surgical repair (a Jahrsdoerfer score of $\geq$6). Nevertheless, approximately one-third of these qualified surgical candidates fail to achieve desirable results, despite their choosing to accept the risks of the complications of atresiaplasty [6].

Up to now, the treatments for CAA had been developed greatly. The Bone-anchored hearing aids (BAHA) system and vibrant sound-bridge (VSB) system are now both widely used in patients with CAA [7,8]. BAHA is also effectively utilized by patients who are not suitable surgical candidates for CAA, being relatively simpler, faster and associated with a lower complication rate [9,10]. These devices raised new challenges and requirements for hearing rehabilitation.

The optimum procedure for hearing rehabilitation in CAA has been controversial for decades. Lambert had proposed that some degradation in hearing outcomes does occur as patients with atresiaplasty are followed beyond the first postoperative year [11]. This finding was confirmed in many other studies [12].

So far there has been only one systematic review that compared the hearing outcomes of atresiaplasty and osseointegrated bone conduction device (OBCD) [12], and one meta-analysis of the complications associated with osseointegrated hearing aids. But there was no meta-analysis of the complications associated with atresiaplasty and long-term hearing outcomes with prolonged follow-up. The aim of the current analysis was thus to review available peer-reviewed literature to examine the overall incidence of complications and the stability of long-term hearing outcomes associated with atresiaplasty.

2. Methods

2.1. Study selection

For the review, five electronic databases (EMBASE, MEDLINE, PubMed, Google Scholar, and Web of Science) were searched to identify titles and abstracts of all possible studies relevant to the topic of CAA and BAHA. All databases were searched from 1994 to 2014. The following terms were used to find eligible studies: aural atresia, microtia atresia, congenital atresia of the external auditory canal, congenital aural malformation, atresiaplasty and congenital auricular atresia. Relevant papers were also reviewed from the reference lists of previous papers for enrollment. Only those papers published in English were selected for the current investigation.

2.2. Data extraction

Two reviewers (C.L.L. & T.Y.Z.) independently screened the database search for titles and abstracts. If either reviewer felt a title and abstract met study eligibility criteria, the full text of the study was retrieved. Two independent reviewers extracted study details pertaining to the inclusion and exclusion criteria using a standardized form. Extracted details included first author, year of publication, follow-up times, number of operated ears, Jahrsdoerfer score, hearing outcomes and complications. Discrepancies were resolved through discussion by the review team.

2.3. Inclusion criteria

The inclusion criteria for this study were as follows: (1) the study population characteristics were patients with CAA; and (2) all studies including one or more of the following outcomes: %ABG $<$30 dB, ΔABG (dB), postoperative ABG (dB) and complications.

2.4. Exclusion criteria

After reviewing the full-text articles, articles were excluded if they were case reports, general review (not systematic review) or commentary, if they only assessed patients with congenital aural stenosis, had less than seven patients in their series. Studies were also excluded from the analysis if they did not include patient outcomes or the outcomes of study were not clearly reported; impossible to extract or calculate the appropriate data from the published results; poor study or reporting quality. When the same institution reported two studies, either the one of better quality or the one of the most recent publication was included unless the study outcomes were mutually exclusive or measured at different intervals. The remaining articles were included.

2.5. Outcomes of interest

We were interested in the following outcomes: (1) Assessment of the long-term hearing outcomes with prolonged follow-up; (2) examine the overall incidence of complications associated with atresiaplasty. For atresiaplasty, we recorded the incidence of postoperative stenosis and bony regrowth, lateralization of TM and facial nerve palsy.

2.6. Statistical analysis

The measure of association used in this meta-analysis was rate with a 95% confidence interval (CI) calculated by Stata 12.0 (College Station, TX, USA). Evidence of heterogeneity was assessed using I^2 and p value, with a fixed-effects model (Mantel–Haenszel method) employed. Publication bias was assessed with Egger's test, Begg's test and funnel plot.

3. Results

3.1. Study selection

Using the above search strategy, 1340 articles were identified on initial search (Fig. 1). Application of the inclusion criteria initially resulted in 37 studies meeting the inclusion criteria. Eighteen studies were excluded after the full text were reviewed, of these 15 studies were excluded for not reporting valuable outcomes and 3 studies were excluded for meeting our exclusion criteria. Finally, 19 studies were included in this meta-analysis, 6 studies were included to evaluate the short-term and long-term hearing outcomes after atresiaplasty.

3.2. Study characteristics

Selected studies were published between 1997 and 2014. The 19 studies covered 964 CAA ears. Sample size for the included

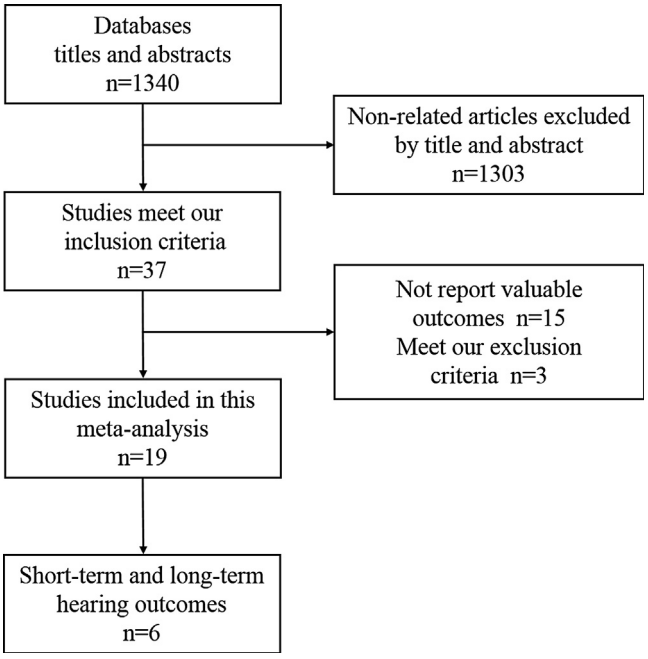

Fig. 1. Summary of study identification and selection.

studies ranged from 8 to 151 patients. Follow-up times ranged from 1 month to 13 years. There were 12 atresiaplasty studies were performed under Jahrsdoerfer scales, of these 1 study was used a modification of the Jahrsdoerfer scale. Especially in recent 5 years, nearly 100% studies used this scale. All 19 studies reported their complications rate. There were 18 studies reported the stenosis and bony regrowth, which occurred in 14.2% (128/900) (range from 0% to 30.3%); 12 studies reported the lateralization of TM, which occurred in 7.5% (53/708) (range from 0% to 18.2%); 15 studies reported the facial nerve palsy, which occurred in 0.5% (4/783) (range from 0% to 5.0%). De la Cruz and Teufert found that the facial nerve was in its normal position in 44 ears (68.8%); it was partially overlapping the footplate in 5 ears (6.6%), and was in an unusual position in 13 ears (17.1%) [13]. The characteristics of all the studies included are summarized in Table 1.

3.3. Meta-analysis

In this review, 6 studies included the short-term (<12 mo) and long-term (>12 mo) hearing outcomes after atresiaplasty [1]. Sample size for these 6 studies ranged from 33 to 107 patients. Some patients loss to follow-up in the long-term studies. But with the time extending, there was a degradation of hearing outcomes in all studies, including the number of ABG < 30 dB, ABG gain and postoperative ABG (Table 2). Digoy and Cueva reported that the number of ABG < 30 dB reduced from 77.8% to 76.9%, but Patel and Shelton reported that reduced from 84.4% to 61.5% [1,14]. The postoperative number of ABG <30 dB between long-term and short-term follow-up were analyzed using meta-analysis with fixed-effect model, there was no significantly heterogeneity ($I^2 = 0\%$, $p = 0.997$), and there was a degradation of hearing outcomes with prolonged follow-up (RR = 1.13, 95%CI: 0.98–1.30) (Fig. 2), meta-analysis with random-effect model was performed with the same results.

A funnel plot of studies included in our outcome of postoperative number of ABG <30 dB was created to explore publication bias (not shown). Effect estimate and confidence intervals were shown on the funnel plot and showed a symmetric distribution around the effect estimate, indicating there was no publication bias in the literature. However only a small number of studies were included in the review, all of which were clinical trials; in this setting, the funnel plot may not truly reflect publication bias and could be misleading and thus was not included in the review.

4. Discussion

This review systematically examined the long-term hearing outcomes and complications of atresiaplasty. Literature search strategies, inclusion and exclusion criteria, data abstraction, and analysis were defined in the protocol. The Jahrsdoerfer grading scale, based on the presence or absence of nine structures, was an extremely important criterion for all surgeons to treat CAA. For atresiaplasty, 12 studies were performed under Jahrsdoerfer scales. For BAHA, Jahrsdoerfer scales do not play an important role in the patient's selection. Sound vibrations were directly transmitted to the skull percutaneously. For these patients with CAA whose Jahrsdoerfer scale < 5, BAHA would be a superior choice.

Table 1
Atresiaplasty articles included.

Reference	Year	Mean follow-up (months)	Jahrsdoerfer score	No. of CAA ears	% Stenosis & bony regrowth	% Lateralization of TM	% Facial nerve palsy
Li et al. [15]	2014	15	≥6	75	10.67	13.3	0
Balaker et al. [16]	2014	9	≥5	55	5.45	–	–
Narushima et al. [17]	2013	17	≥7	8	0	0	0
Memari et al. [18]	2012	12	6.6 ± 1.7	33	30.3	3.0	0
Moon et al. [19]	2012	12	8.32 ± 1.01	98	8.2	–	–
Bouhabel et al. [7]	2012	12	≤9	20	5.0	–	5.0
Siegert [20]	2010	9	[a]	37	5.4	0	0
Yellon [21]	2009	19	≥6	20	20	–	0
Yildirim et al. [22]	2009	27	≥6	9	11.1	–	–
Roberson et al. [23]	2009	4–12	≥6	70	7.1	4.3	0
El-Hoshy et al. [24]	2008	36		40	12.5	5.0	0
Patel and Shelton [14]	2007	>12		64	–	15.6	0
Evans and Kazahaya [25]	2007	65		36	22.2	–	–
Digoy and Cueva [1]	2007	>12		44	6.8	18.2	0
De la Cruz and Teufert [13]	2003	42		116	13.8	3.4	0
Caversaccio et al. [26]	2003	21		18	16.7	–	0
Chang et al. [27]	2002	36	≥6	151	23.8	5.3	1.3
Lambert [11]	1998	33	≥7	50	22	8	1.5 (67 ears)
Murphy et al. [28]	1997	>12		20	20	15	0
Total				964	14.2 (900 ears)	7.5 (708 ears)	0.5 (783 ears)

[a] A modification of the Jahrsdoerfer scale. Only in cases where the patient achieves 20 out of 28 possible points in unilateral and 15/28 in bilateral conditions is middle ear surgery later performed.

Table 2
Short-term and long-term hearing outcomes after atresiaplasty.

Reference	Year	Mean follow-up (months)	No. of CAA ears	No. of ABG <30 dB	ΔABG (dB)	Postoperative ABG (dB)
Memari et al. [18]	2012	2	33	24 (72.7%)	24.7 ± 9.92	28.79 ± 10.23
		12	33	21 (63.6%)	23.3 ± 9.90	30.15 ± 8.88
Moon et al. [19]	2012	3	65	37 (56.9%)	17.39	33.31 ± 11.96
		12	39	19 (48.7%)	17.37	33.33 ± 13.99
Patel and Shelton [14]	2007	3	64	54 (84.4%)	25.1	24.4 ± 8.7
		>12	52	32 (61.5%)	22.8	26.7 ± 11.7
Digoy and Cueva [1]	2007	<12	36	28 (77.8%)	22	23
		>12	13	10 (76.9%)	16	23
Chang et al. [6]	2006	6	100	79 (79%)	23.69	27.16
		36	100	64 (64%)	20.94	29.91
De la Cruz and Teufert [13]	2003	3	107	62 (57.9%)	17.0	28.1 ± 11.7
		42	90	43 (47.8%)	12.9	32.0 ± 13.4

As to assessment of the complications for atresiaplasty, there was heterogeneity among the studies enrolled, which might have been ascribed to the operative numbers and experience, different surgical approach, Jahrsdoerfer scores, skin graft donor sites, postoperative management and follow-up times. Each patient had a unique characteristics and outcomes. Generally, only the most experienced otologist had the ability to do atresiaplasty [20,21]. Only 0.5% patients had the transient facial nerve palsy, all of them recovered within 6 months.

The studies reporting the incidence of complications after atresiaplasty lacks uniformity. Nearly all studies focus on postoperative stenosis and bony regrowth, but some studies neglect lateralization of TM. Lateralization of the TM was a common complication leading hearing loss in CAA, scar tissue contracture within the EAC graft would be one of risk factors [15]. In the correction of atresia, the prevention of complications is more beneficial than treatment. By controlling the risk, the complication rate would be reduced.

The most consistently reported measure of a successful hearing result was a SRT, PTA, or ABG less than 30 dB HL and average hearing gain [12]. All of our six articles included the results of ABG < 30 dB. So we selected it for meta-analysis. In this review, there was a degradation of hearing outcomes with prolonged follow-up (RR = 1.13, 95%CI: 0.98–1.30). The reason might be the status of external aural canal like stenosis, bony regrowth or eczema; the status of tympanic membrane like lateralization, obtuse angle, perforation, granulation or myringitis; the status of ossicular chain. Due to the existence of such degradation, restoring normal or near-normal postoperative hearing would be more challenging. However, we do not know the curve of hearing outcomes, the stability of hearing outcomes need much longer follow-up, 5 years or more.

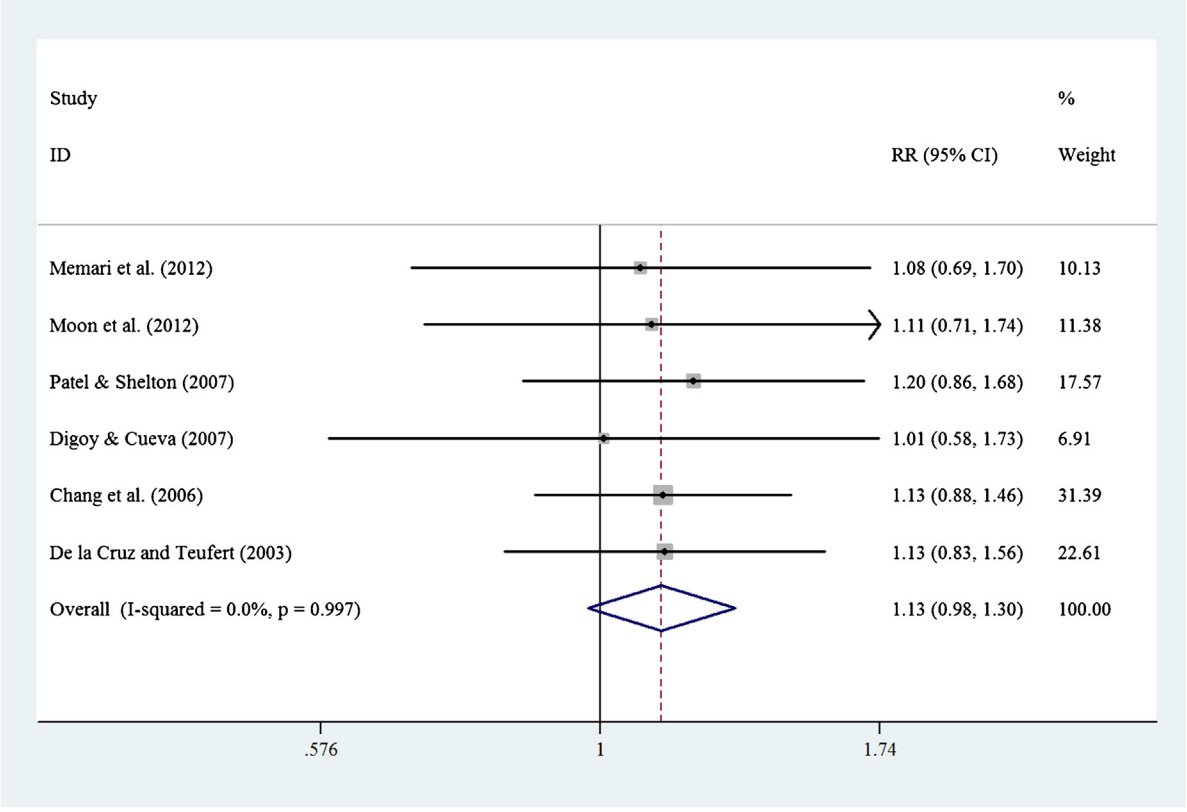

Fig. 2. Forest plot of the risk ratio (RR) for number of ABG <30 dB between long-term and short-term follow-up.

Although BAHA implantation, without consideration of Jahrsdoerfer scale, had more widely applicable indications and excellent hearing outcomes, atresiaplasty had its own advantages. Atresiaplasty could be done combined with microtia surgery [20], and the patients could potentially have a patent and dry aural canal if the surgery was successful. In these successful cases, esthetics could be quite important, especially for the adolescent.

5. Conclusion

The studies reporting the incidence of complications after atresiaplasty lacks uniformity. Nearly all studies focus on postoperative stenosis and bony regrowth, but some studies neglect lateralization of TM. Based on available data, which shows a lack of major complications, such as facial nerve palsy, atresiaplasty is a safe procedure. Even so, our meta-analysis indicated that there was a degradation of hearing outcomes with prolonged follow-up.

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Conflict of interest

The authors declare that they have no conflict of interest.

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