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Outcomes in Pediatric Patients Undergoing Stapedotomy

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ABSTRACT

Objective: Describe outcomes of pediatric stapes surgery at an academic tertiary care center.

Methods: Electronic medical records of patients younger than 21 who underwent stapedotomy between September 2013-July 2020 were reviewed.

Results: 17 patients (7 male, 10 female) were included in our study. 14 underwent surgery on one ear while 3 underwent surgery on both ears (20 ears total). Mean preoperative air-bone gap (ABG) was 34.5 dB (SD 11). At 3 months, the mean postoperative ABG was 20.6 dB (SD 10.2), with a mean improvement of 17 dB (SD 12.1). 64% of patients had closure of their ABG to 20dB or less. A negative correlation between pre-op body-mass index (BMI) and post-op ABG was statistically significant (n=14, p=0.03, r=-0.57 [95% CI -0.85, -0.04]).

Conclusions: Pediatric stapedotomy can be effective and safe. In this cohort, age was not correlated with improvement in ABG; pre-op BMI was significantly correlated with post-op ABG.

KEYWORDS

hearing loss, otology, middle ear surgery, otosclerosis, audiology

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INTRODUCTION

Conductive hearing loss (CHL) in children is most commonly secondary to middle ear effusion; however, a subset of pediatric CHL stems from ossicular chain abnormalities, including juvenile otosclerosis (JO), congenital stapes fixation (CSF), tympanosclerosis, round window atresia, and osteogenesis imperfecta.[1] Stapedectomy using an artificial prosthesis was first done in 1956 and has since been extensively studied in adult patients.[2] In contrast, due to the relative rarity of pediatric ossicular chain pathology, most cohorts that have been studied vis-à-vis stapes surgery are relatively small, although these have been incorporated into systematic reviews and meta-analyses in recent years. Additionally, there has been some hesitancy, historically, to perform stapes surgery on children due to the risk of other congenital abnormalities, like facial nerve anomalies, as well as poorer postoperative outcomes, particularly in patients with CSF.[3] Despite this, recent studies have demonstrated clinically significant reductions of the air-bone gap (ABG) after stapes surgery in CSF and JO patients.[4,5] Schwam et. al found that the presence of concomitant ossicular abnormalities such as malleus fixation did not make a significant difference in outcomes.[6] However, further investigation is warranted given the heterogeneity of available data. In this study, we sought to describe the outcomes for a cohort of pediatric patients who underwent stapes surgery at our institution.

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MATERIALS AND METHODS

IRB approval at Penn State Hershey Medical Center was obtained (STUDY00014969), and eligible patients were identified via the electronic medical record system at Penn State Hershey Medical Center in Hershey, PA. Patients were included in the study if they were younger than 21 years old at the time of surgery and had undergone stapedotomy with perichondral graft and bucket handle prosthesis placement between September 2013-July 2020. Age inclusion criteria encompasses patients who were diagnosed with otosclerosis as children and is in line with other studies of pediatric stapedotomy in the literature.[7,8] All patients were operated on by the same senior author. Demographic data was then collected and organized using Microsoft Excel (version 16.66.1, Redmond, WA). Statistical analyses, including testing for normality, correlations, and t-tests, were performed using Prism 9 (version 9.4.1, Boston, MA).

The following demographic and clinical comorbidities were documented: age, sex, body mass index (BMI), family history of hearing loss, history of chronic otitis media (COM) or mastoiditis, history of otologic procedures (not including previous stapes surgery), history of previous stapes surgery, history of tympanic membrane perforation, and preoperative tinnitus or vertigo. All of these were documented as binary variables, except age and BMI, which were continuous variables. Postoperative complications were collected including sensorineural hearing loss (SNHL), infection, bleeding, persistent vertigo, facial nerve injury, and need for revision surgery.

The audiometric evaluation included preoperative and postoperative audiogram data obtained 3 months following surgery. Variables collected included laterality of ear involved (unilateral vs. bilateral), preoperative and postoperative pure-tone averages (PTA) in air

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conduction and bone conduction, preoperative and postoperative ABG, and successful closure of the ABG. Pure-tone average thresholds were used as a surrogate for overall hearing and were calculated from 0.5, 1, 2, and 3 kHz frequencies. The hearing threshold was calculated by averaging the mean thresholds across all frequencies in the affected ear. ABG was calculated from the air- and bone-conduction thresholds obtained at the same test interval by taking the difference of the bone conduction PTA (PTA-BC) and air conduction PTA (PTA-AC).

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RESULTS AND ANALYSIS

17 patients (7 male, 10 female) were included in our study; of these patients, 14 underwent surgery on one ear while 3 underwent surgery on both ears (20 ears total). Median days of follow-up after surgery, which included ears with audiograms performed beyond 3 months post-op, was 121 (n=18, IQR 279.5). Median age was 16 (n=20, IQR 11) and median BMI was 20.5 (n=20, IQR 10.3). 1 patient had a family history of hearing loss. 8 ears had a history of COM/mastoiditis, 13 had a history of a previous otologic procedure (including 1 which had a previous stapes surgery), and 1 ear had a history of semicircular canal dehiscence syndrome. 1 patient did not have a preoperative or postoperative audiogram and 5 ears did not have a postoperative audiogram at 3 months (i.e., 19 ears with pre-op data available and 14 ears with 3-month post-op data available). Mean preoperative ABG, PTA-AC, PTA-BC were 34.5 dB (SD 11), 52.4 dB (SD 13.2), and 17.8 dB (SD 8.8), respectively. At 3 months, the mean postoperative ABG, PTA-AC, and PTA-BC were 20.6 dB (SD 10.2), 35.4 dB (SD 14), and 16.6 dB (SD 10.4), respectively. Mean improvement in ABG was 17dB (n=14, SD 12.1). Of ears with audiograms done at 3 months post-op, 9 (64%) had a closure of their ABG to 20 dB or less. Age was not significantly correlated with any pre- or post-op audiologic parameters. BMI was significantly correlated with post-op ABG (n=14, p=0.03, r=-0.57 [95% CI -0.85, -0.04]). Patients with previous otologic surgery had a mean ABG improvement of 14.1 (SD 13.0) while those who had no history of otologic surgery had a mean ABG improvement of 20.8 (SD 10.6), though this difference was not statistically significant (p = 0.32). Complications included 1 patient with an intraoperative TM perforation, 1 patient with severe postoperative vertigo and 1 patient requiring revision surgery for adhesions. No patients had profound sensorineural hearing loss in the operative ear after surgery. There were no cases of perilymph gusher or facial nerve injury.

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DISCUSSION

Although stapedotomy is a well-established treatment for CHL related to ossicular chain abnormalities in adults, it is less well-studied in children, given such pathologies are rarer in this population. For instance, one study notes a JO incidence of 0.8 per 100,000 person-years, whereas incidence of otosclerosis in the general population is 3.2 per 100,000 person-years.[9] As a result, much of what is known about managing this entity is built on data compiled from multiple small cohort studies which are then incorporated into larger systematic reviews or meta-analyses. The aim of our study was to add the experiences from our institution to this greater pool of data. A recent meta-analysis of studies that includes 810 ears found a mean pre-op ABG for JO and CSF of 31.8 (SD 5.2) and 39.4 (SD 10), respectively; they found a mean post-op ABG for JO and CSF of 9.6 (SD 6) and 19.2 (SD 12.5), respectively.[4] This is roughly in line with the findings in our cohort. 81% of JO ears and 41% of CSF ears had successful outcomes in that study, which was defined as post-op ABG $<$10 dB.[4] 2 ears in our study (14%) met this criteria. However, other studies have defined post-op ABG of $\leq$20 dB as successful, and 64% of ears in our study met this benchmark.[10,11] Because many of the studies included in Daniel et al. did not include statistical analysis of ABG gain, this was not incorporated into their main results although they did perform a meta-analysis of available data which demonstrated an ABG gain of 24.8 dB for JO and 22.6 dB for CSF which is similar to the findings in our cohort.[4] This improvement can be clinically significant in that it may reduce the need for hearing aids in these patients.

One advantage of our cohort is that we incorporated data on previous otologic surgery; although there was no statistically significant difference in ABG gain between those who did and did not undergo surgery previously, this may be due to the small size of our cohort and suggests

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further investigation into the possibility that this may confound stapes surgery outcomes. In our previous study, which included patients 21 and older, we did find a significantly lower ABG gain in patients who had previous otologic surgery.[12] Another advantage is that we were able to investigate the relationship between history of COM and post-op hearing in our cohort and found no significant difference in post-op audiometric data (PTA-AC, PTA-BC, post-op ABG) between those who did and did not have a history of COM; moreover, none of our patients had post-op SNHL during the follow-up time period of our study. This is reassuring, since previous literature has established that COM increases risk of SNHL and our data suggest that stapes surgery may not impact this risk.[13]

Our finding that higher BMI—which may make for a more difficult procedure given the patient's body habitus—is significantly associated with lower post-op ABG is provocative given our previous study did not find a relationship between BMI and post-op hearing outcomes.[12] Another study, which analyzed BMI as a categorical variable, also did not find any significant impact of this demographic aspect on operative outcomes.[14] That said, neither of the aforementioned studies focused exclusively on pediatric patients. Additionally, prior research has shown that the role of BMI is not as clear cut in predicting tissue adiposity in children as it is in adults.[15] Therefore, further study regarding the impact of BMI on audiometric outcomes of pediatric stapes surgery is warranted.

Our study does have certain limitations, including a small cohort size, limited generalizability—given that all these patients underwent surgery with one provider at one institution, and short follow up (median 121 days). Additionally, we do not have data available to determine the underlying diagnosis that led to stapes surgery in these patients (i.e. JO vs. CSF vs. stapes fixation with other ossicular chain abnormality). Typically, patients with CSF have a more

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significant preoperative hearing loss, undergo surgery at a younger age, and have worse outcomes.[3,4,6] We also do not have data on duration of hearing loss nor on the kind of non-stapes otologic surgery done previously, which may have offered further insight into our cohort's outcomes. Nevertheless, our findings are suggestive of areas that warrant further investigation vis-à-vis pediatric stapedotomy while simultaneously contributing to the growing pool of data on stapedotomy outcomes in this cohort.

In conclusion, pediatric stapedotomy can be effective and safe. Unlike our previously published results, age within the pediatric population did not show correlation to improvement in ABG. Also, in contrast with our previous study, increased BMI was significantly associated with smaller post-op ABG. Patients with previous otologic surgery may have had a worse outcome, although our small sample size was unable to show significance for this.

SUMMARY

- A subset of conductive hearing loss (CHL) in children stems from ossicular chain abnormalities, including juvenile otosclerosis (JO), congenital stapes fixation (CSF), tympanosclerosis, round window atresia, and osteogenesis imperfecta.
- Most cohorts that have been studied vis-à-vis stapes surgery in the pediatric population are relatively small, and there has been some hesitancy historically to perform stapes surgery on children due to risk of concomitant congenital abnormalities such as facial nerve anomalies, or other congenital otologic malformations, as well as poorer postoperative outcomes, particularly with CSF.
- In contrast to previously published results, age within this study cohort did not show correlation to improvement in ABG, and BMI was significantly associated with smaller

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post-op ABG.

- Patients in this cohort with previous otologic surgery may have had a worse outcome, although our small sample size was unable to show significance for this.
- In the realm of pediatric stapes surgery, where reported cases are notably limited, our study adds a meaningful number of cases to the existing pool.

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Competing Interests: The authors declare none.

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aGender (n=17)	Male 7, Female 10
Age (n=20, years)	Median 16 (IQR 8-19)
BMI (n=20, kg/m^2)	Median 20 (IQR 17.3-27)
Family history of hearing loss (n=17)	Yes 1, No 16
History of otologic procedures (n=20)	Yes 13, No 7
History of COM (n=20)	Yes 8, No 12
Laterality of disease (n=17)	Unilateral 10, Bilateral 7
Pre-op PTA-AC (n=19, dB)	Mean 52.4 (SD 13.2)
Pre-op PTA-BC (n=19, dB)	Mean 17.8 (SD 8.8)
Pre-op ABG (n=19, dB)	Mean 34.5 (SD 11)
Post-op PTA-AC (n=14, dB)	Mean 35.4 (SD 14)
Post-op PTA-BC (n=14, dB)	Mean 16.6 (SD 10.4)
Post-op ABG (n=14, dB)	Mean 20.6 (SD 10.2)
Improvement in ABG (n=14, dB)	Mean 17.0 dB (SD 12.1)
Duration of followup (n=18, days)	121 (IQR 80.5-360)

Table I. Demographics of cohort as well as pre- and post-op audiogram findings. BMI=body mass index, IQR=interquartile range, PTA-AC=pure-tone average-air conduction, dB=decibels, PTA-BC=pure-tone average-bone conduction, ABG=air-bone gap

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