



Trans-canal endoscopic ear surgery and canal wall-up tympano-mastoidectomy for pediatric middle ear cholesteatoma

Eran Glikson^{1,2} · Gilad Feinmesser¹ · Doron Sagiv¹ · Michael Wolf^{1,2} · Lela Migirov² · Yisgav Shapira^{1,2}

Received: 17 May 2019 / Accepted: 29 July 2019 / Published online: 3 August 2019
© Springer-Verlag GmbH Germany, part of Springer Nature 2019

Abstract

Purpose To evaluate clinical parameters, outcomes and complications of transcanal endoscopic ear surgeries (EES) and canal wall-up tympano-mastoidectomy (CWU) for middle ear cholesteatoma in children and to compare between the two surgical approaches.

Methods A retrospective chart review of all children (< 16 years) who underwent surgery for cholesteatoma involving the middle ear only with a minimal follow-up period of 12 months. Demographic features, site and extent of disease, outcome and complications were reviewed and compared between the groups.

Results Thirty EES and 19 CWU were included. The overall disease relapse rates in the EES and CWU groups were 20% ($n=6$, residual rate = 10%, recurrence rate = 10%) and 47% ($n=9$, residual rate = 11%, recurrence rate = 37%), respectively ($p=0.04$), with mean duration of follow-up of 32.6 and 37.2 months, respectively. In the EES and CWU groups, the most common site of residual disease was the mastoid cavity/antrum ($n=2$, 66% and $n=2$, 100%, respectively). Most recurrences involved the epitympanum and extended into the tympanic cavity ($n=2$, 66%) in the EES group and into the tympanic cavity, posterior mesotympanum and mastoid cavity/antrum ($n=3$, 43%, each) in the CWU group. The overall complication rates in the EES and CWU groups were 10% ($n=3$) and 11% ($n=2$), respectively ($p=0.61$).

Conclusions Endoscopic ear surgeries in children were found to be an acceptable and safe technique for the treatment of cholesteatoma limited to the middle ear cavity. A better overall success rate and a similar complication rate were found in the EES group when compared to CWU.

Keywords Cholesteatoma · Endoscopic ear surgery · Pediatric · Canal wall up · Mastoidectomy

Introduction

The annual incidence of cholesteatoma in pediatric patients is reported to be 3–6 per 100,000 [1]. Cholesteatoma in children is more aggressive and has a higher recurrence rate as compared to adults [2]. The greater degree of upper airway infections, and otitis media caused by Eustachian tube dysfunction, is probably promoting factors for cholesteatoma development in the pediatric population.

Surgical management of pediatric cholesteatoma is currently controversial. Traditional treatment options include

canal wall-down (CWD) or canal wall-up (CWU) tympano-mastoidectomy. The overall reported rates of residual cholesteatoma in all age groups reach 7% (range 2–21%) and 22% (range 15–27%) for CWD and CWU mastoidectomies, respectively, and the recurrence rates reach 5% (range 0–10%) and 25% (range 9–70%), respectively [3–6]. Previous reports suggest an increased incidence of residual disease in children (up to 51%) [1, 2].

Since operative endoscopy was introduced into ear surgery, it has significantly changed surgical, anatomic and physiologic concepts [7]. The endoscopes provide better visualization of common sites of residual cholesteatoma and may, therefore, decrease residual disease, enabling preservation of the ear canal wall. Transcanal endoscopic tympano-atticotomy (EES) is indicated for cholesteatoma confined to the tympanic cavity and attic without involving the mastoid [8].

✉ Eran Glikson
Eran.Glikson@sheba.health.gov.il

¹ Department of Otolaryngology, Head and Neck Surgery, Sheba Medical Center, 5262100 Tel-Hashomer, Israel

² Sackler Faculty of Medicine, Tel Aviv University, Tel Aviv, Israel

Early reports of EES for cholesteatoma failed to differentiate children from adults [9–13] with only a few studies describing EES results in the pediatric population [14–16].

The current study presents our experience with trans-canal endoscopic ear surgery and CWU tympano-mastoidectomies for cholesteatoma limited to the middle ear in pediatric patients.

Materials and methods

We retrospectively reviewed all children aged < 16 years who underwent surgery for middle ear cholesteatoma between March 2009 and March 2016, in the Department of Otolaryngology and Head and Neck Surgery at the Sheba Medical Center—a tertiary referral center, with a minimal follow-up period of 12 months.

The study includes two study groups:

1. Children who underwent trans-canal endoscopic or endoscopy-assisted tympano-atticotomy (EES).
2. Children who underwent CWU for cholesteatoma and the disease was found to be limited to the middle ear.

This ensured a similar disease extent between the groups, thus making a comparison possible. All cases in both groups were performed by the same three surgeons (L.M, M.W and Y.S).

The decision to perform mastoidectomy vs EES was influenced by several factors. In the first years of the study period as EES was only introduced in our practice, most surgeries were done using microscopy. Also before our routine use of non-echo-planar diffusion-weighted magnetic resonance imaging (non-EPI DWI MRI), the information regarding disease extent into the mastoid was more limited and, therefore, mastoidectomy was chosen.

Canal wall-up tympano-mastoidectomy was defined when exclusive microscopic mastoidectomy was performed with the preservation of the posterior canal wall. These cases were selected to undergo mastoidectomy and the decision to perform CWU technique was made during the operation when the disease extent was limited to the middle ear and attic.

Transcanal endoscopic surgery was indicated when the cholesteatoma did not extend posterior to the lateral semicircular canal (LSCC).

Rigid endoscopes (Hopkins Telescope, Karl Storz) 3 mm in diameter, 0°, 30°, 45°, and 70°, were used routinely for all the procedures with a three-chip video camera (Karl Storz) and 20-inch high-resolution monitor.

Illumination was provided by a xenon cold light source connected to a fiber-optic light cable with average light intensity of 50%. A special set of micro-endoscopic instruments

[angled picks, suction and forceps (Karl Storz)] was used in addition to the routine otologic microinstruments.

All surgeries were performed under general anesthesia. Facial nerve monitoring was used in all surgeries.

Preoperative assessment included high-resolution computed tomography (CT) of the temporal bones and/or non-echo-planar diffusion-weighted magnetic resonance imaging (non-EPI DWI MRI) and pure-tone audiometry.

Our standard post-operative follow-up recommendations include repeated clinical examinations (at 2 weeks and at 2, 6, and 12 months after surgery), audiologic assessment 2–4 months post-surgery and performance of non-EPI DWI MRI at approximately 1 year post-surgery. MRI is then performed annually/bi-annually.

Recurrent cholesteatoma was defined as a lesion arising in a new retraction pocket diagnosed otoscopically. Residual disease was defined as cholesteatoma diagnosed by non-EPI DWI MRI behind an intact tympanic membrane.

Complications were divided into major and minor. Minor complications were defined as those resolving spontaneously or with conservative therapy. Major complications were defined as those necessitating revision surgeries or resulting in permanent sequelae. We further divided the complications as those that occurred intraoperatively or post-operatively.

The study was reviewed and approved by the Research Ethics Committee of the Chaim Sheba Medical Center, Tel-Hashomer, Israel (application number: 2581-15-SMC).

Statistical analysis

Continuous variables are displayed as mean \pm standard deviation and categorical variables are presented as number and percent in each group.

The mean and standard deviation of each measurement were determined. Chi-square test, Fisher's exact test and Student's *T* test were used to compare continuous and categorical variables between the groups. All calculations were considered significant at $p < 0.05$ (two tailed).

Results

A total of 45 surgeries for cholesteatoma were performed in children with the use of an endoscope, of which there were 34 operations described as trans-canal. Four children were excluded due to a short post-operative follow-up (< 12 months). Altogether, 30 operations (28 patients) were included in the study group.

A total of 81 mastoidectomies for cholesteatoma were performed in children, of which 28 children underwent CWU mastoidectomy for cholesteatoma. The cholesteatoma was found to be limited to the middle ear in 19 surgeries.

The main characteristics of the study groups are listed in Table 1. Both groups showed similar characteristics. The mean patient ages in the EES and CWU groups were 11.1 years (range 3–16) and 10.6 years (range 6–16), respectively. In both groups, approximately 60% of cases underwent primary surgery while the rest were revision cases.

Cholesteatoma extension

The sites of cholesteatoma involvement in the EES are noted in Fig. 1. The most common site of involvement was the posterior epitympanum ($n=23$, 85%), followed by the anterior epitympanum ($n=8$, 30%), posterior mesotympanum (defined as the area along the posterior wall of the middle ear lateral to the pyramidal eminence) ($n=7$, 26%) and sinus tympani (defined as the area along the posterior wall of the middle ear medial to the pyramidal eminence) ($n=7$, 26%).

Facial nerve dehiscence and involvement of the chorda tympani nerve were noted in six (20%) and seven (23%) cases, respectively.

In the CWU group, the most common sites of cholesteatoma involvement were the posterior epitympanum ($n=14$, 74%), anterior epitympanum ($n=10$, 52%), sinus tympani ($n=2$, 11%) and hypotympanum ($n=2$, 11%).

Complications

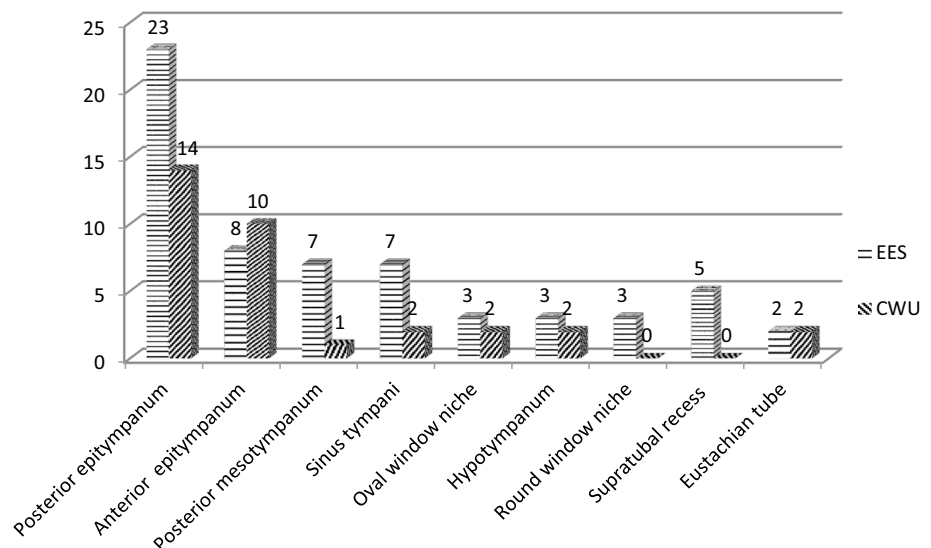
The overall complication rates in the EES group and the CWU groups were 10% ($n=3$) and 11% ($n=2$), respectively ($p=0.61$). In the EES group, two minor and one major complications were noted including one transient post-operative vertigo, one surgical site seroma that was drained on post-operative day 4 and one case of labyrinthitis that resulted in permanent moderate–severe hearing loss.

Table 1 Characteristics of children undergoing ear surgery for cholesteatoma

	EES, <i>n</i> (%)	CWU, <i>n</i> (%)	<i>p</i> value
Total	28 (30 operations)	18 (19 operations)	
Male	16 (57%)	19 (72%)	0.58
Female	12 (43%)	5 (28%)	
Primary surgery	18 (60%)	11 (58%)	0.82
Revision surgery	12 (40%)	8 (42%)	
EES	2	0	
Microscopic atticotomy	4	1	
Mastoidectomy	6	7	
Mean age (years)	11.11 (range 3–16)	10.6 (range 6–16)	0.38
Affected side			
Right	10 (33%)	8 (42%)	0.64
Left	20 (67%)	11 (58%)	

EES transcanal endoscopic ear surgery, CWU canal wall up tympanomastoidectomy

Fig. 1 Sites of cholesteatoma involvement



In the CWU group, one minor and one major complications were noted including one intra-operative dislocation of the stapes during the removal of cholesteatoma which resulted in transient vertigo, one surgical site-infected hematoma, and one case of post-operative permanent moderate hearing loss.

Follow-up

The mean durations of follow-up in the EES and CWU groups were 32.6 and 37.2 months, respectively.

The overall disease relapse rates in the EES and CWU groups were 20% ($n=6$) and 43% ($n=12$), respectively ($p=0.04$). The rates of disease recurrence in the EES and CWU groups were 10% ($n=3$) and 36.8% ($n=7$), respectively ($p=0.02$). The surgical complications and outcome including the residual and recurrence rates are summarized in Table 2.

The sites of residua and recurrence are listed in Table 3. In the EES and CWU groups, the most common site of residual disease was the mastoid cavity/antrum ($n=2$, 66% and $n=2$, 100%, respectively). In the EES group, most recurrences involved the epitympanum and extended into the tympanic cavity ($n=2$, 66%). In the CWU group, most recurrences extended from the epitympanum into the tympanic cavity, posterior mesotympanum and mastoid cavity/antrum ($n=3$, 43%, each).

Five of six EES patients with recurrence/residual underwent radical/modified radical mastoidectomy, and one patient with an epitympanic recurrence underwent revision EES.

Hearing evaluation

The mean pre-operative air conduction pure tone average (AC-PTA) in the EES group was 37.2 dB. A total of 21 patients (75%) presented with hearing loss (HL) of any degree, 19 were pure conductive HL and the rest were mixed type. The mean word recognition score (WRS) was 92%.

The mean post-operative AC-PTA was 39.6 dB and the mean post-operative WRS was 84.4%. These did not differ significantly from the pre-operative scores.

In the CWU group, the mean pre-operative AC PTA and WRS were 41.5 dB and 90%, respectively. The mean post-operative AC PTA and WRS were 42.2 dB and 88%, respectively. These did not differ significantly from the pre-operative scores.

Discussion

Although cholesteatoma surgery is being performed worldwide for decades, it continues to pose a challenge. The choice among well-described surgical techniques depends on disease extension, surgeon's own experience, published data, and the institution's resources.

Pediatric cholesteatoma has been described as an aggressive disease with higher recidivism rates compared to adult patients [1, 2, 16], in some extent due to the limited exposure and visualization of the entire middle ear spaces when using microscopic vision.

Table 3 Cholesteatoma residual and recurrence sites

	EES, <i>n</i> (%)	CWU, <i>n</i> (%)
Residual		
Total	3	2
Mastoid antrum/cavity	2 (66.6%)	2 (100%)
Oval window niche	1 (33.3%)	0
Tympanic cavity	1 (33.3%)	1 (50%)
Posterior mesotympanum ^a	1 (33.3%)	1 (50%)
Sinus tympani	0	1 (50%)
Recurrence		
Total	3	7
Epitympanum	3 (100%)	7 (100%)
Tympanic cavity	2 (66.6%)	3 (43%)
Posterior mesotympanum ^a	0	3 (43%)
Sinus tympani	0	2 (28%)
Mastoid antrum/cavity	0	3 (43%)
Oval window niche	0	2 (28%)

^aDefined as the area along the posterior wall of the middle ear lateral to the pyramidal eminence

Table 2 Surgical complications and outcome

	EES, <i>n</i> (%)	CWU, <i>n</i> (%)	<i>p</i> value
Complications	3 (10%)	2 (11%)	0.6
Minor	2 (6.7%)	1 (5.5%)	
Major	1 (3.3%)	1 (5.5%)	
Disease relapse	6 (20%)	9 (47%)	0.04
Residual	3 (10%)	2 (10.6%)	0.7
Recurrence	3 (10%)	7 (36.8%)	0.02
Follow-up period (months)	32.6 (range 12–65)	37.2 (range 12–74)	

The ear endoscope's main advantage is improved visualization, especially in previously difficult to access locations such as the posterior mesotympanum (facial recess), sinus tympani, anterior epitympanic space, and the Eustachian tube [8–12]. All of these can be observed through a transcanal approach in many cases. The main advantages of a transcanal approach are preservation of the external canal's posterior wall, thus preserving its structure and function, and preservation of the mastoid bone leading to a decrease of possible intraoperative complications such as dural injury and intracranial complications. An intact canal wall approach, when feasible, is generally favored for pediatric cholesteatoma surgery [17].

According to our experience, other advantages of transcanal EES are less painful surgery and shorter healing time compared with traditional surgeries [18].

In this study, the results of transcanal EES for middle ear cholesteatoma in pediatrics are described and compared to a group of children with similar disease extent who underwent CWU tympano-mastoidectomy (the relevant traditional microscopic surgical alternative). Other surgical alternatives to address middle ear cholesteatoma using a microscope include middle ear exploration and transcanal atticotomy with or without retrograde mastoidectomy.

Our study group of EES patients showed a relatively high rate of cholesteatoma involvement of the anterior epitympanic space, sinus tympani, and posterior mesotympanum, which are all sites of potential residual disease—classically considered inaccessible by transcanal approach. However, cholesteatoma was accessed and removed from the above-mentioned sites. The prevalence of involved sites in this study is similar to a previous study performed in our institution, which included adult patients who underwent transcanal endoscopic ear surgeries for cholesteatoma [18].

Early EES cohorts presented the results in adults and reported cholesteatoma residual and recurrence rates between 0 and 15.5% and 0 and 8.2%, respectively [9–13]. In our previous study of EES in adults (performed by the same surgeons as the current study) [18], the residual and recurrence rates were 10% and 8.3%, respectively. In recent years, a few studies focused on EES in children and reported residual and recurrence rates between 10.5 and 20% and 10.3 and 13.2%, respectively [14–16]. In this study, the recurrence and residual rates of the endoscopic surgery group are slightly below the lower end of this range.

The EES study group was compared to a CWU group who were operated by the same surgeons. EES' overall recurrence/residual rate was found to be lower than CWU mastoidectomy (20% vs 47%, $p=0.04$). The difference was noted in the recurrence rate (10% vs 36.8%, $p=0.02$) and not in residual disease rates. We hypothesize that the reasons may be better visualization of the Eustachian tube patency when using an endoscope compared to retroauricular

mastoidectomy. When the anterior tympanic membrane is intact, it is difficult to visualize and inspect the Eustachian tube when using a microscope, whereas the endoscope provides perfect visualization of the orifice. Another reason may be less formation of epitympanic adhesions when performing the endoscopic approach due to improved visualization and lack of drilling.

Data regarding complications in EES cases are insufficient. Previous studies in adults provide little or no information regarding this crucial issue. Some authors describe briefly that no complications related to the use of the endoscope [9] or that no intraoperative facial nerve injury [11] was found. Other authors do not address the issue of complications [10, 13]. In our previous study of EES in adults, we reported overall minor and major complication rates of 16.6% and 6%, respectively [18].

The few previous studies of pediatric EES provided limited information regarding complications. Hunter et al. reported that no complications were found [15]. Marchioni et al. reported 3 complications in a combined group of 59 EES and CWU mastoidectomies [14] and Ghadersohi et al. reported 2 minor surgical site complications (3%) [16].

The minor and major complication rates of pediatric EES in this study were 6.7% and 3.3%, respectively. These results were similar to the CWU group and are lower than the reported complication rates of EES in adults [18].

Previous reports of endoscopic surgeries and the current study are all limited by their cohort size and follow-up period, which are related to the recent introduction of endoscopic middle ear surgery. Other limitations of this study are its retrospective design and possible selection bias. Though a similarly dated, consecutive patient sample who underwent CWU tympano-mastoidectomies for a disease with a similar extent by the same surgeons acted as a control group, a selection bias is still possible.

Despite these limitations, we present a large series of transcanal EES and a control group of CWU mastoidectomies in pediatrics, and report detailed disease description, surgical results and residual/recurrence sites.

Conclusions

Transcanal endoscopic ear surgery in pediatrics was found to be an acceptable and safe technique for the treatment of select cases in which cholesteatoma was limited to the middle ear cavity.

A better overall success rate and a similar complication rate were found in the EES group when compared to CWU.

Funding None.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval The study was reviewed and approved by the Research Ethics Committee of the Chaim Sheba Medical Center, Tel-Hashomer, Israel (application number: 2581-15-SMC).

References

1. Sohet JA, De Jong AL (2002) The management of pediatric cholesteatoma. *Otolaryngol Clin N Am* 35:841–851
2. Glasscock ME III, Dickins JR, Wiet R (1981) Cholesteatoma in children. *Laryngoscope* 91:1743–1753
3. de Zinis LO, Tonni D, Barezani MG (2010) Single-stage canal wall-down tympanoplasty: long-term results and prognostic factors. *Ann Otol Rhinol Laryngol* 119:304–312
4. Haginomori S, Takamaki A, Nonaka R et al (2008) Residual cholesteatoma: incidence and localization in canal wall down tympanoplasty with soft-wall reconstruction. *Arch Otolaryngol Head Neck Surg* 134:652–657
5. Sanna M, Facharzt AA, Russo A et al (2009) Modified Bondy's technique: refinements of the surgical technique and long-term results. *Otol Neurotol* 30:64–69
6. Tomlin J, Chang D, McCutcheon B et al (2013) Surgical technique and recurrence in cholesteatoma: a meta-analysis. *Audiol Neurootol* 18:135–142
7. Thomassin JM, Duchon-Doris JM, Emram B et al (1990) Endoscopic ear surgery. Initial evaluation. *Ann Otolaryngol Chir Cervicofac* 107:564–570
8. Migirov L, Shapira Y, Horowitz Z et al (2011) Exclusive endoscopic ear surgery for acquired cholesteatoma: preliminary results. *Otol Neurotol* 32:433–436
9. Badr-El-Dine M (2002) Value of ear endoscopy in cholesteatoma surgery. *Otol Neurotol* 23:631–635
10. Ayache S, Tramier B, Strunski V (2008) Otoendoscopy in cholesteatoma surgery of the middle ear. What benefits can be expected? *Otol Neurotol* 29:1085–1090
11. Tarabichi M (2004) Endoscopic management of limited attic cholesteatoma. *Laryngoscope* 114:1157–1162
12. Presutti L, Gioacchini FM, Alicandri-Ciufelli M et al (2014) Results of endoscopic middle ear surgery for cholesteatoma treatment: a systematic review. *Acta Otorhinolaryngol Ital* 34(3):153–157
13. Marchioni D, Villari D, Mattioli F et al (2013) Endoscopic management of attic cholesteatoma: a single-institution experience. *Otolaryngol Clin N Am* 46:201–209
14. Marchioni D, Soloperto D, Rubini A et al (2015) Endoscopic exclusive transcanal approach to the tympanic cavity cholesteatoma in pediatric patients: our experience. *Int J Pediatr Otorhinolaryngol* 79:316–322
15. Hunter JB, Zuniga MG, Sweeney AD et al (2016) Pediatric endoscopic cholesteatoma surgery. *Otolaryngol Head Neck Surg* 154(6):1121–1127
16. Ghadersohi S, Carter JM, Hoff SR (2017) Endoscopic transcanal approach to the middle ear for management of pediatric cholesteatoma. *Laryngoscope*. 127:2653–2658
17. Osborn AJ, Papsin BC, James AL (2012) Clinical indications for canal wall-down mastoidectomy in a pediatric population. *Otolaryngol Head Neck Surg* 147:316–322
18. Glikson E, Yousovich R, Mansour J, Wolf M, Migirov L, Shapira Y (2017) Transcanal endoscopic ear surgery for middle ear cholesteatoma. *Otol Neurotol*. 38(5):e41–e45

Publisher's Note Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.