OTOLOGY



Efficiency of intraoperative endoscopic inspection in reducing residuals in canal-wall-up surgery for pediatric cholesteatoma involving the mastoid

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Abstract

Objective To investigate the efficiency of additional intraoperative endoscopic inspection in reducing residual cholesteatoma in pediatric cholesteatoma involving the mastoid treated with classic canal-wall-up mastoidectomy and tympanoplasty.

Materials and methods 32 cases of pediatric cholesteatoma involving the mastoid were enrolled in this perspective study and treated with classic canal-wall-up mastoidectomy and tympanoplasty. Transmastoid posterior tympanotomy, atticotomy and transecting tendon of tympani tensor were conducted to achieve adequate visualization of hidden spaces in the middle ear. After complete removal of cholesteatoma, endoscopic inspection was additionally performed to check residual cholesteatoma. All cases had at least a 2-year follow-up by routine otoscopy examination, CT scan or MR imaging. Residual rates of both intraoperative and follow-up findings were used to evaluate the efficiency of the endoscopic inspection in reducing residual cholesteatoma and compared with published reports.

Results The additional intraoperative endoscopic inspection did not find any residual in this case series. In the 2-year followup, 2 cases (2/32, 6.3%) with residual cholesteatoma and 3 cases with recurrence (3/32, 9.4%) were found. The mean duration of endoscopic inspection and microscopic procedure were 17.9 min and 93.6 min, respectively.

Conclusions This study suggested that the additional intraoperative endoscopic inspection in microscopic CWU surgery for pediatric cholesteatoma involving the mastoid had no obvious value in reducing residual cholesteatoma but took extra time.

Keywords Cholesteatoma · Endoscopy · Residual cholesteatoma · Canal-wall-up mastoidectomy, CWU · The mastoid

Introduction

Surgical management of pediatric cholesteatoma is challenging because of its characteristics of aggressive growth in the well-pneumatized middle ear and high possibilities of residual and recurrence [1–4]. In recent years, endoscopy has been used more and more in middle ear surgery. Compared with microsurgery, endoscopic surgery provided improved visualization of hidden spaces in middle ear cavity and reduced invasion [5]. Previous study suggested that

² NHC Key Laboratory of Hearing Medicine (Fudan University), Shanghai 200031, China involvement of endoscopy resulted in a low rate of mastoidectomy in pediatric cholesteatoma [6]. For cholesteatoma limited in middle ear, the required procedures could be accomplished by exclusive transcanal endoscopic approach. However, for that extended into the mastoid, mastoidectomy was usually necessary to sufficiently expose the mastoid air cells for complete removal of cholesteatoma, especially for cases with well-pneumatized mastoid. For these cases, exclusive transcanal endoscopic surgery is usually incapable to completely remove diseases in the mastoid if posterior canal wall was kept intact. Therefore, traditional microscopic canal-wall-up (CWU) mastoidectomy was thought to be necessary in this situation by most surgeons [7]. However, after complete removal of cholesteatoma by microscopic CWU procedure, the necessity of additional intraoperative endoscopic inspection for reducing residuals has not yet reached a consensus. The effect of assistant or exclusive endoscopic surgery on reducing residuals of pediatric middle ear cholesteatoma has been evaluated in several studies. It was

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shown that residual cholesteatoma was found by adjunctive endoscopic inspections in 16-38% cases after the traditional microscopic surgeries [8–10]. In two recent reviews with meta-analysis, pediatric cholesteatoma treated by endoscope dissection was shown to be associated with lower residual rate than that treated by microscopic dissection with or without endoscope inspection [6, 8]. However, to our best knowledge, no individual previous report suggested that there was statistically significant difference between residual rate of pediatric cholesteatoma treated by microscopic surgery and that treated by exclusive or assistant endoscopic procedures. In addition, the enrolled cases in most of the previous reports had different extents of cholesteatoma and were treated with different microscopic procedures, such as transcanal atticotomy, CWU or canal-wall-down (CWD) tympanomastoidectomy. Therefore, it is still unclear whether additional intraoperative endoscopic inspection is necessary for reducing residual rate after classic CWU tympanomastoidectomy for pediatric cholesteatoma involving the mastoid.

In this study, we enrolled 32 cases of pediatric cholesteatoma involving the mastoid. After classic CWU tympanomastoidectomy, an additional endoscopic inspection was performed to find the residual cholesteatoma. The residual rates determined in the first operation and follow-up were compared to reported residual rates in reviewed previous studies to evaluate value of addition endoscopic inspection in the mentioned circumstance.

Methods

Study design

This study is a monocenter perspective single-arm study in a tertiary hospital. The protocol of the investigation was approved by institutional ethics committee.

Patients

From March 2017 to September 2020, 32 cases of pediatric middle ear cholesteatoma with the mastoid involved were enrolled in this study. The inclusion criteria were: (1) ages under 16 years; (2) CT scans indicating low-density signals in the well-pneumatized mastoid; (3) cholesteatoma extended beyond the posterior dome of the lateral semicircular canal, which confirmed in the operation; (4) no history of ear surgery. All patients had CT scans and pure-tone audiometry (PTA) or auditory steady-state response (ASSR) before the operation. The diagnosis of middle ear cholesteatoma was based on conductive or mixed hearing loss with or without ear perforation, low-density signal in the middle ear, and bone erosion of ossicular chain on CT images. The etiologies of cholesteatomas were defined as congenital or acquired according to the presence of an intact tympanic membrane or not, respectively. All cases had at least 2-year postoperative follow-ups.

Surgical procedure

CWU tympanomastoidectomy was conducted when lowdensity signal blocked the antrum and was presented in the mastoid on preoperative CT. Mastoidectomy with intact auditory canal wall was performed through postauricular incision. Transmastoid posterior tympanotomy, atticotomy and transecting tendon of tympani tensor were conducted to achieve adequate visualization of hidden spaces in the middle ear. To avoid missing cholesteatoma matrix, the removal of cholesteatoma was completed using capsule-tracking technology, i.e., dissecting cholesteatoma by continuously tracking its capsule wall. Bone drilling and cholesteatoma removal were alternately performed step by step to avoid unintentional destruction of cholesteatoma capsule and leaving matrix in deep air cells. In cases that cholesteatoma had no intact capsules, it was removed as much as possible before bone drilling and irritation. The tensor tympani tendon was transected for an improved visualization of anterior tympanum. When the cholesteatoma was believed to be completely removed under microscopy, endoscopy was used to inspect the middle ear for any residual cholesteatoma through external auditory canal, the mastoid, and the attic. The inspection was sequentially conducted in the attic, supratubal recess, anterior tympanic fold, tensor tympani tendon, eustachian tube, the stapes, tympanic sinus, facial recess, and hypotympanum. Ossicular reconstruction and repair of tympanic membrane were then performed if necessary.

Follow-up

Routine follow-ups were at 6 months, 1 year and 2 years after the operations. Examination of tympanic membrane was performed at each follow-up and CT scans were performed every year. Diffusion-weighted magnetic resonance imaging (DWI) was applied if CT images of the middle ear and the mastoid had low-density signals suspected to be cholesteatoma. Secondary surgeries were conducted in cases with new retractions or pockets in tympanic membrane or with suspected cholesteatoma indicated by DWI MR imaging. Cholesteatoma confirmed in the operation was defined as residual or recurrence by whether tympanic membrane was intact or not, respectively.

Statistics

Residual rate revealed by endoscopic inspection in the first operation was calculated by subtracting the number of

cases detected with residuals from the total number of cases. Residual rate in follow-up was subtraction of the number of cases with cholesteatoma and intact tympanic membrane from the total number of cases. Analysis was performed using GraphPad Prism 9.3.0. The residual rates were compared with those reported in the previous studies by Chisquare test or Fisher's exact test.

Results

Residual cholesteatoma found by additional endoscopic inspection

A total of 32 pediatric cholesteatoma involving the mastoid were resected by microscopic CWU tympanomastoidectomy and checked for residual by additional endoscopic inspection. Seventeen cases were diagnosed as congenital cholesteatoma and 15 cases were diagnosed as acquired. The classification of procedures in this study is EES 1 [11]. During resecting cholesteatoma in microscopic procedure, 1 case was found to have cholesteatoma in deep tympanic sinus, which was out of direct microscopic visualization. The hidden part of cholesteatoma was then removed under 30° endoscopy. For all the other 31 cases, endoscopic inspection after the microscopic CWU procedures did not find any residual cholesteatoma.

Residual rate in the follow-up

In the first year after operation, two cases were found to have recurrent cholesteatoma, no case having residual. In the second year of follow-up, one case of recurrent cholesteatoma and two cases of residual ones were found. The overall residual rate in the 2-year follow-up was 6.3% (2/32), while the recurrent rate was 9.4% (3/32, Table 1). One residual was from acquired cholesteatoma and found in the protympanum. The other one was from congenital cholesteatoma and found in mesotympanum which likely originated from remnants on the medial side of tympanic membrane. All the three cases with recurrence had retractive pocket at pars flaccid of tympanic membrane. Of them, two cases were from acquired cholesteatoma and one case was from congenital one.

Table 1 Residual and recurrence in follow-up

| | Congenital | Acquired | Total | Rate |
|-------------|------------|----------|-------|------|
| Case number | 21 | 11 | 32 | |
| Residual | 2 | 0 | 2 | 6.3% |
| Recurrence | 0 | 3 | 3 | 9.4% |

Operation time

The operation time from the incision to closure and the time for endoscopic inspection were recorded. Endoscopic time was determined as the duration between shifting the microscopy away and back. The mean duration of endoscopic inspection was 17.9 min, while that of microscopic procedure was 93.6 min.

Discussion

In this study, we used not only postoperative findings of residual cholesteatoma in 2-year follow-up but also intraoperative findings by additional endoscopic inspection to evaluate its necessity after classic CWU procedure in pediatric cholesteatoma involving the mastoid. If the additional endoscopic inspection could find extra cholesteatoma after microscopic dissection, it would be valuable in reducing residual cholesteatoma.

There were two previous studies that specifically reported residual rates in pediatric cholesteatoma involving the mastoid and treated with microscopic CWU tympanomastoidectomy (EES 0) [12, 13]. Their pooled residual rate was 24.1% (32/101) which was significantly higher than that of the present study (P = 0.0167, one-sided Fisher's exact test, Fig. 1). To the best of our knowledge, there was no report presenting specific residual data of pediatric cholesteatoma involving the mastoid and treated with microscopic CWU plus endoscopic inspection (EES 1). Three previous studies reported residual rates of EES 1-treated pediatric

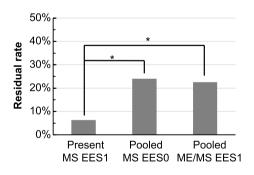


Fig. 1 Comparison of the present residual rate with that in published reports with the same disease extension or surgical technique. *Legend*: The pooled residual rate in published reports of pediatric cholesteatoma involving the mastoid and treated with EES 0 was significantly higher than that of the present study treated with EES 1 (P = 0.0167). The pooled residual rate from published reports on EES 1-treated pediatric cholesteatoma including both that limited in the middle ear and that involving the mastoid was also significantly higher than that in the present study (P = 0.0229). One-sided Fisher's exact test. *EES 0* microscopic CWU, *EES 1* microscopic CWU with following endoscopic inspection, *CWU* microscopic canal-wall-up mastoidectomy and tympanoplasty

cholesteatoma including both that limited in the middle ear and that involving the mastoid [14–16]. The pooled residual rate from three previous reports was 22. 6% (37/164) which was also significantly higher than that in the present study (P=0.0229, one-sided Fisher's exact test, Fig. 1).

To our best knowledge, this study is the first report on the effect of additional endoscopic inspection after microscopic CWU surgery on reducing residuals of pediatric cholesteatoma involving the mastoid. Several studies have investigated the effect of endoscopy application in pediatric cholesteatoma surgery [7, 12, 14–17]. However, each of these reports failed to find a statistically significant reduction of residual rates in pediatric cholesteatoma after using adjunctive endoscopic inspection or dissection, or even exclusive endoscopic

Statistically Author Extent of EES classification Cases (n) Surgical technol-Residual cases Residual rate Investigative cholesteafactors significance year ogy toma Yaniv ME or MS EES₀ 42 CWU 16 38.1% Endoscopic No 2019 49 CWU and EI 9 inspection EES 1 18.4% With reduced drilling amount Glikson ME EES 0 19 CWU 2 10.5% Microscopic or No 2019 endoscopic EES 3 30 TEES 3 10.0% surgery MS EES₀ 28 CWU 10 34.4% Microscopic or Marchioni No 2015 endoscopic ME EES 3 31 TEES 6 19.3% surgery 47 CWU 4 Hunter ME or MS EES 0 8.5% 8.5% Microscopic or No 2016 endoscopic 21 10.3% MS EES 2 Combined M-E 2 9.5% surgery surgery 8 EES 3 TEES 12.5% ME 1 Ghadersohi ME or MS 7 CWU and EI 2 28.5% 28.5% EES 1 Endoscopic No 2017 inspection or 9 MS EES 2 Combined M-E 2 22.2% 6.5% dissection surgery ME EES 3 22 TEES 0 0.0% ME or MS 108 CWU and EI James EES 1 26 24.1% Endoscopic No 2016 With reduced inspection or drilling amount dissection EES 2 127 Combined M-E 19 17.0% surgery With reduced drilling amount Dixon ME EES 0, 1 64 With or without 7 10.9% Exclusive endo-No 2020 CWU and EI scopic surgery EES 3 6.3% 64 TEES 4 ME or MS CWU with or Cohen EES 0, 1 24 6 25.0% Microscopic or No 2017 without EI endoscopic dissection 32 7 EES 2, 3 Combined M-E 21.9% surgery or TEES 7 Sarcu ME or MS EES 1, 2 42 With or without 16.7% N.A N.A 2015 atticotomy and CWU Intraoperative endoscopic inspection 105 Piras MS EES 0 CWU 22 21.0% N.A N.A 2021

Table 2 Review of previous studies investigating the impact of different surgical technologies on residual rate of pediatric cholesteatoma

ME cholesteatoma limited in the middle ear, *MS* cholesteatoma involving the mastoid, *EES* endoscopic ear surgery, *EES* 0 microscopic CWU, *EES1* microscopic CWU with following endoscopic inspection, *EES* 2 combined endoscopy- and microscopy-guided surgery, *EES* 3 transcanal exclusive endoscopic surgery (TEES), *CWU* microscopic canal-wall-up mastoidectomy and tympanoplasty, *EI* endoscopic inspection, *M-E* microscopy- and endoscopy-guided surgery, *N.A.* not applicable

surgery (Table 2). Recently, a meta-analysis study suggested that transcanal exclusive endoscopy surgery (TEES) had a significant lower residual rate than microscopic surgery in pediatric cholesteatoma [6]. Another meta-analysis study also demonstrated that pediatric cholesteatoma treated with TEES brought about reduced residual and recurrence rates of TEES when compared to that treated with the traditional microscopic surgery [8]. However, it should be noted that the extent of cholesteatoma and microscopic surgical technique were not considered in the studies mentioned above. Actually, most of the previous reports did not investigate endoscopy effect, respectively, in cases of pediatric cholesteatoma limited in the middle ear and that extended to the mastoid (Table 2). There were only two reports with clearly defined cholesteatoma extent which were limited in the middle ear without involvement of the mastoid. For these cases, TEES and microscopic CWU surgery had comparable residual rates around 10% [18, 19]. For other reports on cholesteatoma without clearly defined extents, the residual rates of surgeries with different degree of endoscopy application varied from 8.5 to 38.1%, but their difference in each individual report still had no statistical significance (Table 3, Fig. 2) [7, 10, 12, 14–17]. The difference of cholesteatoma extent might contribute to the variations of residual rates in those reports. Cholesteatoma limited in the middle ear could be removed by traditional microscopic CWU surgery or TEES, while that extended into the mastoid was usually treated by microscopic tympanomastoidectomy. In fact, some previous reports compared cases treated with different surgical techniques that were chosen according to the extent of cholesteatoma [7, 12, 18]. The selection bias might affect the results of comparisons among different EES techniques.

Our case series only included pediatric cholesteatoma involving the mastoid which required for mastoidectomy to remove diseases in the mastoid. The additional intraoperative endoscopic inspection did not find any left matrix after microscopic cholesteatoma resection. This result suggested that there was no obvious value of additional intraoperative endoscopic inspection in detecting residual cholesteatoma after classic CWU surgery. In contrast with our study, a previous study found 7 in 42 cases (16.7%) having left cholesteatoma matrix after microscopic surgery [10]; however, this report included both cholesteatomas limited in the middle ear and that extended into the mastoid. Additionally, not all cases had CWU tympanomastoidectomy. All cases in our study had mastoidectomy, transecting tendon of tympani tensor, transmastoid atticotomy, and posterior tympanotomy, which could provide an improved visualization in most of the hidden space of the middle ear. The persist applications of these procedures and capsule-tracking technology could reduce left matrix after microscopy-guided resection of cholesteatoma.

Endoscopy undoubtedly had its value in the surgical treatment of pediatric cholesteatoma, especially for examining and resecting diseases in some extraordinarily hidden area. As shown in this study, one case had cholesteatoma in hidden space, which was finally removed under angled endoscopy. Capsule tracking technology allowed the surgeon aware of these hidden diseases. If they were removed by microscopic dissection, more bone removal would be required for sufficient visualization, while the application of angled endoscopy might reduce bone work and possible injury to important structures, such as facial nerve and the footplate.

As shown in this study, residual cholesteatoma was still found in the case series during the follow-up even after additional intraoperative endoscopic inspection. However, the residual rate of our EES 1 cases was much lower compared to that in pooled EES 0 cases with the mastoid involvement in the previous reports. The residual rates of cholesteatoma after similar surgical techniques also varied a lot in the published reports [13]. The variation of residual rates might be due to the difference of surgical techniques and experiences from different person and affiliation. The reduction of residual rate might also partly attribute to the techniques of capsule-tracking and transection of tympani tensor tendon. In the previous reports of EES 1 cases including both that limited in the middle ear and extend into the mastoid, the pooled residual rates were higher than that in this study. The inclusion of cases with cholesteatoma limited in the middle ear might contribute to the increase of residual rate. In addition, it should be pointed out that some surgeons might reduce the amount of bone drilling in EES 1 surgery, because following endoscopic inspection could provide an adequate visualization in this circumstance. However, the reduction of bone removal might interrupt tracking of cholesteatoma matrix and leave matrix in extraordinarily hidden space or unopened air cells.

The additional endoscopic inspection unsurprisingly increased the duration of operation in this study. Some reported EES 1 surgery with reduced bone drilling amount might save a little time when compared to classic microscopic CWU surgeries. However, additional inspection time would be much longer than the saved drilling time. Therefore, EES 0 CWU surgery might still take less time than EES 1 CWU surgery.

In summary, for pediatric cholesteatoma involving the mastoid and treated with CWU surgery, additional endoscopic inspection after microscopic dissection had no obvious value in detecting residual cholesteatoma. Although endoscopy had obvious value in dissecting cholesteatoma of extraordinarily hidden spaces, routine additional endoscopic inspection for residual cholesteatoma

| | Juigical weiling | Surgical technology Author, year | Data of ind | Data of individual report | | Pooled data | а | | Statistical analysis Total cases Residual cases Residual | Total cases | Residual cases | Residual |
|--------------------|------------------|----------------------------------|-------------|---------------------------|----------------------|-------------|------------------|------------------------|--|-------------|----------------|----------|
| cholestea- toma | | | Total cases | Residual cases | Residual rate (%) | Total cases | s Residual cases | s Residual rate (%) | | | | rate (%) |
| ME | EES 0 | Glikson, 2019 | 19 | 2 | 10.5 | 19 | 2 | 10.5 | Chi-square | 238 | 23 | 9.7 |
| | EES 3 | Glikson, 2019 | 30 | ŝ | 10.0 | 155 | 14 | 9.0 | DF: 0.206, 2 | | | |
| | | Hunter, 2016 | 8 | 1 | 12.5 | | | | P = 0.902 | | | |
| | | Ghadersohi, 2017 | 22 | 0 | 0.0 | | | | | | | |
| | | Dixon, 2020 | 64 | 4 | 6.3 | | | | | | | |
| | | Marchioni, 2015 | 31 | 9 | 19.3 | | | | | | | |
| | EES 0, 1 | Dixon, 2020 | 64 | 7 | 10.9 | 64 | 7 | 10.9 | | | | |
| MS | EES 0 | Piras, 2021 | 105 | 22 | 21.0 | 133 | 32 | 24.1 | Chi-square | 163 | 36 | 22.1 |
| | | Marchioni, 2015 | 28 | 10 | 34.4 | | | | DF: 1.637, 1 | | | |
| | EES 2 | Hunter, 2016 | 21 | 2 | 9.5 | 30 | 4 | 13.3 | P = 0.200 | | | |
| | | Ghadersohi, 2017 | 6 | 2 | 22.2 | | | | | | | |
| ME or MS | EES 0 | Yaniv, 2019 | 42 | 16 | 38.1 | 89 | 20 | 22.5 | Chi-square | 477 | 96 | 20.1 |
| | | Hunter, 2016 | 47 | 4 | 8.5 | | | | DF: 3.811, 5 | | | |
| | EES 1 | Yaniv, 2019 | 49 | 6 | 18.4 | 164 | 37 | 22.6 | 1/C.0 = A | | | |
| | | Ghadersohi, 2017 | 7 | 2 | 28.5 | | | | | | | |
| | | James, 2016 | 108 | 26 | 24.1 | | | | | | | |
| | EES 2 | James, 2016 | 127 | 19 | 17.0 | 127 | 19 | 17.0 | | | | |
| | EES 0, 1 | Cohen, 2017 | 24 | 9 | 25.0 | 24 | 9 | 25.0 | | | | |
| | EES 1, 2 | Sarcu, 2015 | 42 | 7 | 16.7 | 42 | 7 | 16.7 | | | | |
| | EES 2, 3 | Cohen, 2017 | 32 | 7 | 21.9 | 32 | 7 | 21.9 | | | | |

 Table 3
 Summarized residual data grouped by different cholesteatoma extents and surgical technologies in the previous reports

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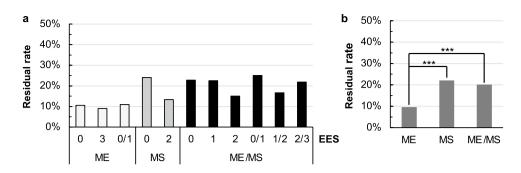


Fig. 2 Comparisons of residual rates in pediatric cholesteatomas with different disease extensions and surgical techniques in previous reports. *Legend*: **a** The residual rates of cases treated with different surgical techniques were comparable when grouped by disease extension as ME, MS and ME/MS. **b** Regardless of surgical technique, there was significantly statistical difference among the residual rates of pediatric cholesteatoma with different extensions. The pooled residual rates of cases with cholesteatoma extended into the mastoid

after complete microscopic CWU surgery with adequate visualization had no obvious necessity. Endoscopy is a useful tool but could not naturally get a better outcome in surgery of pediatric cholesteatoma without a comprehensive understanding of the disease and the surgery. In addition, the usage of endoscopy in pediatric cholesteatoma involving the mastoid should be optional but not mandatory, especially after complete microscopic CWU surgery.

Data Availability All data analysed during this study are available from the corresponding author on reasonable request.

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(MS or ME/MS) were higher than that of cases with cholesteatoma limited in the middle ear (ME). ***, P < 0.001, two-sided Fisher's exact test. *ME* cholesteatoma limited in the middle ear, *MS* cholesteatoma involving the mastoid, *EES 0* microscopic CWU, *EES 1* microscopic CWU with following endoscopic inspection, *EES 2* combined endoscopy- and microscopy-guided surgery, *EES 3* transcanal exclusive endoscopic surgery

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