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Original Article

Endoscopic Ear Surgery for Congenital Cholesteatoma in Children

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BACKGROUND: Endoscopic ear surgery is a promising technique for removing congenital cholesteatoma in children. It can provide greater visual access to hidden areas of the middle ear and facilitate middle-ear manipulation. This study compares a single-center experience in managing congenital cholesteatoma with an endoscopic approach with that in managing congenital cholesteatoma with a conventional microscopic approach.

METHODS: Records of consecutive patients aged under 8 with congenital cholesteatoma confined to the middle ear at our tertiary referral hospital from January 2013 to December 2018 were retrospectively reviewed. Operation time, hospital stay, postoperative complications, and recurrence/residue of congenital cholesteatoma were compared between patients receiving microscopic versus endoscopic surgery.

RESULTS: A total of 33 pediatric patients aged from 19 months to 7 years were enrolled; 12 children underwent microscopic surgery, and 21 received an endoscopic approach for removing congenital cholesteatoma. The mean operative time was 1.61 hours for the microscopic group and 1.49 hours for the endoscopic group without statistical difference. No postoperative sensorineural hearing loss and complications were reported. Four cases of recurrence/residue were observed on the follow-up endoscopic exam or computed tomography, and no differences were shown between the 2 groups. Of the total patients, 94.7% (n = 11) in the microscopic group and 90.5% (n = 19) in the endoscopic group demonstrated an intact tympanic membrane without perforation or retraction after surgery. No audiological differences were reported between the 2 groups.

CONCLUSION: Endoscopic ear surgery can effectively and safely remove congenital cholesteatoma in children and is not inferior to conventional microscopic approaches.

KEYWORDS: Congenital cholesteatoma, endoscopic ear surgery, EES

INTRODUCTION

Congenital cholesteatoma (CC) presents as a whitish mass with keratinized squamous epithelium behind an intact tympanic membrane (TM) and is most prevalent in young patients.[1] In pediatric patients, the annual incidence of CC is reported to be 3-6 per 100 000. Congenital cholesteatoma is approximately 1.4 times more prevalent in males than in females.[3,4] Recently, early diagnosis of CC has become prevalent due to growing interest in otitis media in pediatric patients, widespread application of otoscopy and endoscopy, and increased use of audiometric screening systems.[5] In addition, the routine use of computed tomography (CT) imaging has also increased the detection rate of early-stage CC.[6]

The pathogenesis of cholesteatoma is not thoroughly understood. Congenital cholesteatoma is thought to be a squamous cell growth with origins in the epibranchial placode or Michael's body, leading to cystic lesion formation adjacent to the tensor tympani tendon.[7]

Early surgical removal is of clinical importance since the growth of CC is highly correlated with patient age.[8] However, even with early intervention, some risk of residual or recurrent CC remains. Residual cholesteatoma results from the incomplete surgical removal of the cholesteatoma matrix, while recurrent cholesteatoma results from the reformation of the retraction pocket after a complete

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previous surgical cholesteatoma removal.[9] Complete removal of CC is essential; however, previous studies show that the proportions of residual and recurrent cholesteatoma are variable.[10-13] Ossicular chain involvement is a risk factor for recurrent disease,[14] and stage, type (open or closed), and location also affect rates of residual and recurrent CC. Controlling CC in difficult or hidden areas is necessary for reducing recurrent and residual CC. These hidden areas include the sinus tympani, facial recess, anterior epitympanic space, and hypotympanum in the middle ear.

It is important to thoroughly dissect attachment sites such as the malleus and tensor tympani tendon (ASMT) or cochleariform process, which are common origin sites of CC, for complete removal.[15] However, it is difficult to examine these areas thoroughly with conventional microscopic surgery.[16] Endoscopic surgery is now applied in otology and is a feasible option for managing CC, particularly in the pediatric population.[17-19] Endoscopic surgical instruments provide a minimally invasive approach with a wide surgical view.[16] The conventional microscope provides a magnified image with excellent quality; however, only a limited, narrow segment of the external auditory canal is exposed, whereas the endoscope provides wider angles of view.[20] Therefore, the endoscopic approach can provide greater visual access to hidden areas of the middle ear and facilitate middle-ear manipulation.

Endoscopic approaches for ear surgery have come to prominence recently and represent one of the most promising techniques for removing CC. Here, a single-center experience in managing CC is reported, comparing an endoscopic approach to the conventional microscopic approach in terms of surgical outcomes.

MATERIALS AND METHODS

Study Design and Patients

Electronic medical records were used to identify patients who were diagnosed with CC at our tertiary referral hospital from January 2013 to December 2018. Patients aged under 8 years with clinically and pathologically proven CC were included. Among those, CC only confined in the middle ear that represented as Potsic's staging system[21] from stages 1 to 3 were enrolled. All cholesteatomas were limited in tympanum without attic extension. Congenital cholesteatoma was clinically diagnosed from the finding of a whitish mass visible through an intact tympanic membrane preoperatively and a mass confined in the middle ear intraoperatively. Pathological findings of the mass confirmed the diagnosis of cholesteatoma.

A total of 33 pediatric patients aged from 19 months to 7 years were included in this study. All patients underwent temporal bone computed tomography (TBCT) to clarify the characteristics and extent of CC. Patients then underwent microscopic or endoscopic CC removal, performed by a single surgeon. Patients who underwent CC removal under a microscopic view were allocated to the microscopic group (n = 12), and those who underwent surgery under endoscopic view were allocated to the endoscopic group (n = 21). Patients were allocated in chronological order. Microscopic approach was performed during previous half of the study period, and endoscopic approach was performed during last half of the study period. In the microscopic group, 6 out of 12 cases were performed using endaural incision with a widening of the orifice with a 2-prong retractor, whereas 6 cases were performed using only transcanal incision (Lempert type 1) and an elliptical speculum sized 3.0 × 5.0 mm or 4.0 × 6.0 mm to access the middle ear cavity. All 21 cases in the endoscopic group underwent transcanal incision only (Figure 1). The microscopic group was operated with a microscope, exclusively, while endoscopic groups were operated with an endoscope, exclusively without hybrid methods. The optimal goal of all surgical cases was to eradicate all visible CC, leaving no residual CC. Once visible CCs were removed, microscopic or endoscopic examinations were performed to examine the middle-ear cavity for possible residual CC. In endoscopic cases, both 0° and 30° angled endoscopies, with diameter sized 3.0 mm, were used to maximize visualization. Most of the small-sized closed-type cholesteatoma was removed without disruption of the cyst wall by appropriate handling with suction and other instruments. In case of big size cholesteatoma, it was difficult to remove in one piece; therefore, first an incision was made in the matrix part, the internal keratin material was removed, and then the remnant matrix, including cystic wall, was removed.

Patients were followed up at an outpatient clinic every 3-6 months, and after 1 year, tympanic membrane examination and TBCT were performed.

This retrospective study protocol was reviewed and approved by the Institutional Review Board of Asan Medical Center (2019-0579), and informed consent was waived.

Variables

Patient demographic and clinical data were collected, including age, sex, mean follow-up period, side, size, and location of CC, Potsic's staging system,[21] CC type (closed/open), and presence of preoperative eroded ossicles. The size of cholesteatoma was measured mostly after the cholesteatoma was removed during surgery. If it was not done, pathology report was used. In case of open type, we measured the size of cholesteatoma when removed from middle ear and used the CT finding with reference. "Closed type" refers to the congenital cholesteatoma with an intact cystic membrane, whereas "open type" refers to the congenital cholesteatoma with ruptured cystic membrane. Eroded ossicles were predicted by preoperative CT and confirmed by intraoperative endoscopic exam. For surgical outcomes, operation time; from the start to the end of the surgical procedure, hospital stay after surgery, postoperative complications, residual or recurrent CC, postoperative sensorineural hearing loss, postoperative tympanic membrane status, and audiological differences were evaluated and compared.

Measurement of Auditory Function

Auditory function was determined by pure tone audiometry (PTA), and mean hearing levels were expressed as the average of hearing thresholds at 500, 1000, and 2000 Hz (3-frequency average, 3FA) and 500, 1000, 2000, and 4000 Hz (weighted 4-frequency average, W4FA). In the audiology evaluation, all ossiculoplasty cases were excluded as the effect of hearing gain from ossiculoplasty was ruled out. Most of the patients were too young to be evaluated using PTA. Therefore, only 4 patients in the microscopic group and 5 patients in the endoscopic group underwent both preoperative and postoperative audiology tests.

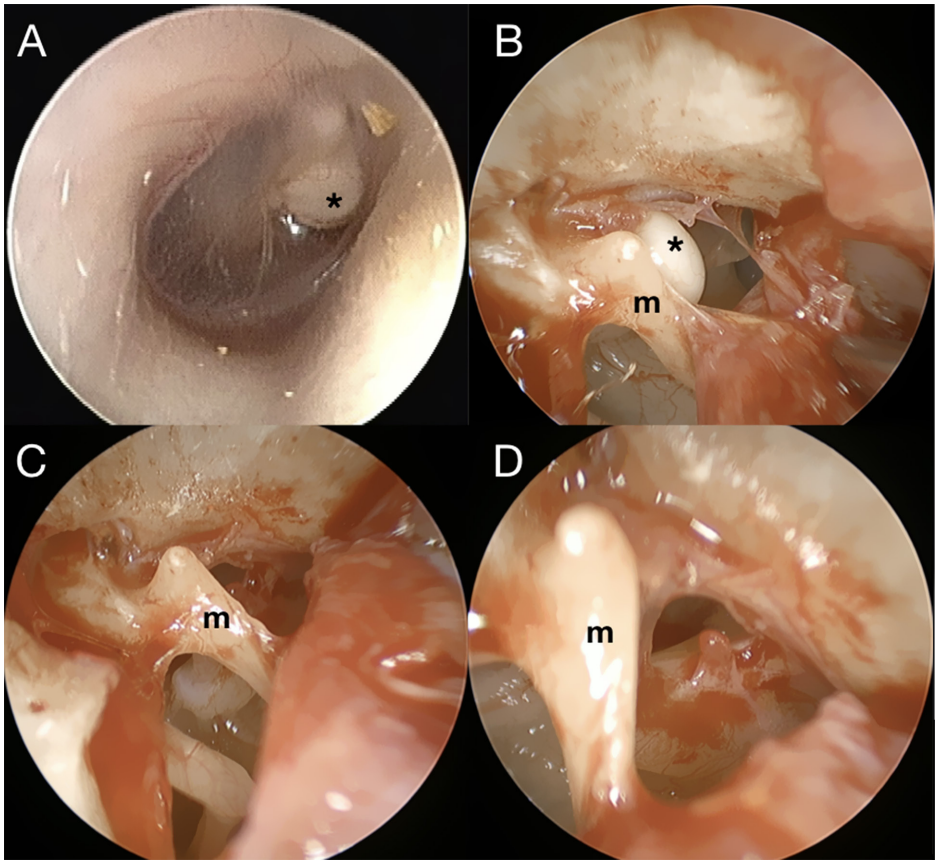

Figure 1. a-d. The surgical procedure for congenital cholesteatoma via the endoscopic approach. (a) A tympanomeatal flap was elevated with transcanal incision; (b) keratin pearl was observed anterior to the umbo and cholesteatoma was removed; (c) no visible mucosal lesion or cholesteatoma sac was identified under endoscopic exam; (d) the tympanomeal flap was repositioned.

Statistical Analysis

Continuous variables were expressed as mean and standard deviation (or standard error) or median and range, while categorical variables were expressed as number and percentage. Variables were compared using a *t*-test, chi-squared test, or Fisher's exact test. A *P* value of less than.05 was considered to indicate statistical significance. Statistical analyses were performed using IBM SPSS software, version 24.0 (IBM SPSS Corp.; Armonk, NY, USA).

RESULTS

The study included 33 patients, 22 males and 11 females, with a median age of 3.43 (±1.20) years. Patient demographic data are summarized in Table 1. Patients in the endoscopic group (3.10 ± 0.95) were statistically younger than those in the endoscopic group (4.00 ± 1.41) (*P* = .036). There was no dominant side for CC between the endoscopic and microscopic groups (*P* = .692). The median CC size was 5.2 (± 1.8) mm, range from 2.5 to 8.5 mm, and 3.2 (± 1.7) mm, range from 1.5 to 7 mm, in the microscopic and endoscopic groups, respectively, and it demonstrated that the median CC size in the microscopic group is statistically bigger than the endoscopic group (*P* = .004). The most dominant location of CC was anterosuperior quadrant (ASQ) in both groups, but there were no differences between the 2 groups (*P* = .861). Potsic's stage 1 was reported for 63.7% of cases and no difference between the 2 groups was observed (*P* = .446). The closed type of CC was more frequent than the open type (69.7% vs. 30.3%), but rates were not different between the 2 groups. One case of open-type cholesteatoma in the microscopic group was located in the ASQ. In endoscopic group, 4 was in anterosuperior, 1 in anteroinferior, 1 in posteroinferior, and 3 in posterosuperior quadrant. There were also no differences between the 2 groups in terms of the preoperative presence of eroded ossicles (*P* = .686). Only one from the endoscopic group underwent ossiculoplasty, otherwise ossiculoplasty was not performed in the rest of the patients with eroded ossicles.

The mean follow-up time was significantly different between the 2 groups at 43.97 months for the microscopic group and 22.02 months for the endoscopic group (*P* = .010).

The mean operation time for the endoscopic group was 19.8 minutes longer than for the microscopic group, 1.16 hours for the microscopic group and 1.49 hours for the endoscopic group, respectively, but this difference was not statistically significant (*P* = .107). Endoscopic surgery was associated with a 0.15 days shorter mean hospital stay compared to microscopic surgery, 1.25 days for the microscopic group, and 1.10 days for the endoscopic group, respectively, but this difference was also not statistically significant (*P* = .508).

Not all patients underwent PTA testing due to lack of test cooperability in younger patients. When comparing the improvement of hearing levels measured by 3FA and W4FA, there were no significant differences between microscopic and endoscopic groups (*P* = .305,

Table 1. Patients Characteristics

Variable	Total (n = 33)	Microscopic Group (n =12)	Endoscopic Group (n = 21)	*P*
Age (y), mean (SD)	3.43 (1.20)	4.00 (1.41)	3.10 (0.95)	.036
Sex (%)				.249
Male	22 (66.7)	10 (83.3)	12 (57.1)	
Female	11 (33.3)	2 (16.7)	9 (42.9)	
Mean follow-up	30.00 (20.31)	43.97 (23.63)	22.02 (13.00)	.010
Side of cholesteatoma (%)				.692
Left	15 (45.5)	6 (50.0)	9 (42.9)	
Right	18 (54.5)	6 (50.0)	12 (57.1)	
Size of cholesteatoma (mm)	3.9 (2.0)	5.2 (1.8)	3.2 (1.7)	.004
Location of CC*				.861
ASQ	20 (60.6)	8 (66.7)	12 (57.1)	
AIQ	2 (6.1)	1 (8.3)	1 (4.8)	
PSQ	8 (24.2)	2 (16.7)	6 (28.6)	
PIQ	3 (9.1)	1 (8.3)	2 (9.5)	
Potsic's staging system				.446
Stage 1	23 (69.7)	7 (58.3)	16 (76.2)	
Stage 2	3 (9.1)	2 (16.7)	1 (4.8)	
Stage 3	7 (21.2)	3 (25.0)	4 (19.0)	
Cholesteatoma type				.054
Closed	23 (69.7)	11 (91.7)	12 (57.1)	
Open	10 (30.3)	1 (8.3)	9 (42.9)	
Preoperative eroded ossicles	7 (21.2)	3 (25.0)	4 (19.0)	.686

Variables are expressed as number (percentage) unless indicated otherwise.
*Location of CC was determined by the most dominant quadrant that CC occupied.
CC, congenital cholesteatoma; ASQ, anterosuperior quadrant; AIQ, Anterior inferior quadrant; PSQ, Posterior superior quadrant; PIQ, Posterior superior quadrant.

P = .456). 3FA difference was −2.50 dB in the microscopic group and 3.67 dB in the endoscopic group, and the W4FA difference was −1.67 dB in the microscopic group and 2.17 dB in the endoscopic group.

Postoperative PTA was also conducted for patients with stage 1 or 2 CC by Potsic's system with intact ossicles (Figure 2). Eight patients in the microscopic group and 12 patients in the endoscopic group were evaluated. Pure tone audiometry demonstrated a slight difference in hearing level both in 3FA and W4FA at 1.88dB and 1.74dB, respectively, but this was not statistically significant between microscopic and endoscopic groups (*P* = .728, *P* = .750).

DISCUSSION

Pediatric CC demonstrates aggressive clinical features compared to adult forms, with higher recurrence rates.[19] Congenital cholesteatoma may involve the pars tensa and can consequently spread to the sinus tympani, facial recess, and epitympanum, which are poorly visualized regions.[22] Visualization is particularly challenging in children, who constitute the majority of CC patients. Early detection is essential for the cure of CC, and awareness of CC has increased owing to the use of otoendoscopy in local clinics.[5] If CC is detected late, there is a risk of posterior extension associated with poor prognosis. Ossicular erosion and mastoid invasion are also risk factors for poor prognosis of CC.[21,23] Complete surgical removal is essential for successful management of CC. For complete resection of CC, common sites of attachment, including the ASMT region, should be dissected.[24] With conventional microscopic approaches, thorough visualization of these hidden areas is challenging, owing to the working angle of the microscope. In one study, a Buckingham mirror was used for microscopic surgery to investigate the blind spot in the middle ear; however, resolution of the surgical field was poor.[15] In these circumstances, the introduction of endoscopy has conferred significant benefits in the removal of CCs.

The current study describes a single-center experience in managing CC via an endoscopic approach and shows the feasibility of endoscopic compared with microscopic surgery in otology. This study compares postoperative outcomes between 2 groups receiving conventional microscopic or endoscopic surgery for CC, which were balanced in terms of baseline patient demographic data, except age, and size of CC. It represented that patients who underwent surgery via endoscopic approach who were even younger resulted in comparable surgical outcome to patients who underwent surgery via microscopic approach. All surgeries in the microscopic and endoscopic group were performed by a single surgeon, minimizing the chances of different surgical outcomes as a result of differences in surgical skill. Also in this study, no hybrid technique was performed, only microscopic or endoscopic approach was performed. Moreover, without extension to mastoid cavity, all cases were performed without postauricular incision. No additional approach was performed once the trans-canal approach was decided according to

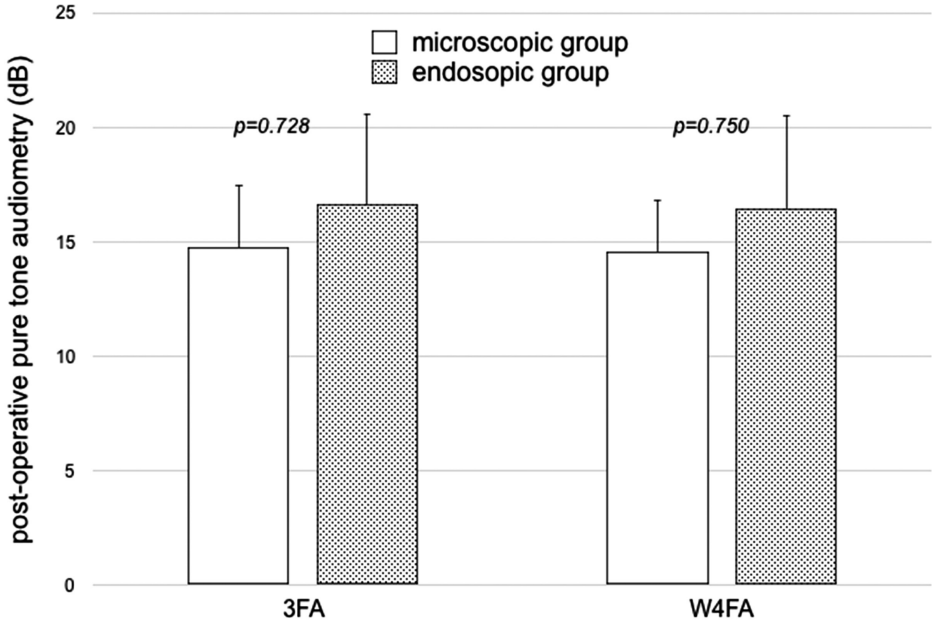

Figure 2. Post-operative pure tone audiometry of patients with stage 1 or 2 CC by Potsic's system with intact ossicles. There was no statistically significant difference in postoperative 3FA and W4FA between microscopic and endoscopic groups. (*P* value = .728 in 3FA and .750 in W4FA)

pre-operative decision-making with examining the TBCT. Based on TBCT, the decision-making process may focus on whether the post-auricular approach is needed or not. Congenital cholesteatoma confined in the middle ear could be removed through the microscopic or endoscopic approach, solely. Only endaural incision was made when ear canal is too small to use speculum alone, in case of microscopic group. All cases were decided either trans-canal or postauricular approach based on TBCT finding, and none of them changed from one to another. If cholesteatoma was extended to mastoid cavity, we decided that the trans-canal approach is not enough. In case of endoscopic approach, it was not impossible to perform endoscopic only surgery for CC confined in middle ear. A previous study also demonstrated that in pediatric patients who have narrow external auditory canal, tympanoplasty via endoscopic surgery was successfully performed. In that study, the smallest anterior-posterior diameter of the canal was 3.2 mm, while the smallest superior-inferior diameter was 3.4 mm.[25] In case of a very narrow canal, a device that widens the canal might be helpful.[26]

Even though our study only took the microscopic or endoscopic approach solely, a hybrid technique could be performed if cholesteatoma is too huge to be visualized through microscope. There is a study that a hybrid technique, mainly performed in a microscope but assisted by an endoscope, was useful in acquired attic cholesteatoma.[27] However, choosing one of many techniques (microscopic, endoscopic, hybrid) would be dependent on the surgeon's preference supported by the facility and experience.

This study showed no significant differences in surgical outcomes for CC between the microscopic and endoscopic surgery groups, although median follow-up periods were significantly different between groups. The reason for the significant difference in the median follow-up period between the 2 groups is explained by the later advent of endoscopy compared to microscopy to the otology field. In recent years, the practice has changed from the microscopic approach to an endoscopic approach for CC removal. Hence, before the introduction of endoscopy, all transcanal CC removals were conducted only by microscopy. Because all the earlier patients were allocated to the microscopic group rather than the endoscopic group, there was a significant follow-up difference between the 2 groups. Though it was not statistically significant, this study demonstrated a somewhat longer operation time for the endoscopic than the microscopic group. This may have arisen due to the recent introduction of endoscopic surgery, with the requirement to adapt to the new technology. This difference might become smaller as endoscopic approaches become more widespread for transcanal CC removal.

A shorter postoperative hospital stay was observed for the endoscopic group than for the microscopic group, although this was not statistically significant. This may relate to shorter recovery times for the endoscopic group. All patients were discharged when pain was tolerably controlled by oral medication, and acute postoperative complications were not observed. In transcanal endoscopic approaches, endaural incision and canaloplasty are unnecessary, whereas these techniques are frequently applied in microscopic approaches for wider visualization. Minimal manipulation of the external auditory canal in endoscopic surgery results in reduced postoperative discomfort, a possible explanation for the shorter hospital stay after surgery, but further investigation of immediate postoperative pain, subjective discomfort, and other postoperative factors are needed to support this theory.

Four recurrence/residue cases, 2 in each group, were reported, and no differences were observed. Those cholesteatomas were found 33.75 months after the initial surgery. Even cholesteatomas were found several months after initial surgery, all of them were found to be in the same quadrant of the original disease. Therefore, it is ambiguous to distinguish residual from recurrence in those cases. Even immediate postoperative examination showed no residual lesion. Chances are that microscopic residual remained.

Table 2. Surgical Outcomes

Variable	Total (n = 33)	Microscopic Group (n = 12)	Endoscopic Group (n = 21)	*P*
Postoperative complication (%)	0	0	0	NA
Residue (%)	0	0	0	NA
Recurrence (%)	4 (12.1)	2 (16.7)	2 (9.5)	.610
Postoperative SNHL (%)	0	0	0	NA
Postoperative TM status (%)				.147
Normal	30 (90.9)	11 (91.7)	19 (90.5)	
Attic retraction	2 (6.1)	0	2 (9.5)	
Posterior retraction	1 (3.0)	1 (8.3)	0	

SNHL, Sensorineural hearing loss; TM, Tympanic membrane.

Table 3. Surgical Outcomes for Potsic's System Stage 1

Variable	Total (n = 23)	Microscopic Group (n = 7)	Endoscopic Group (n = 16)	*P*
Postoperative complication (%)	0	0	0	NA
Recurrence (%)	2 (8.7)	2 (28.6)	0	.083
Postoperative TM status (%)				NA
Normal	23 (100)	7 (100)	16 (100)	
Attic retraction	0	0	0	
Posterior retraction	0	0	0	
Operation time (hours)	1.17 ± .0.27	1.05 ± 0.25	1.23 ± 0.26	.132
Hospital day after surgery (day)	1.00 ± 0.30	1.00 ± 0.58	1.00 ± 0.00	1.000

This study describes the successful use of endoscopic ear surgery for CC in young patients and demonstrates the strength of the endoscopic approach in terms of clinical outcomes. There are several advantages to the endoscopic approach in diseases of the middle-ear cavity in general. Firstly, endoscopic approach provides a wider visual field than conventional microscopy and allows detailed examination of the middle ear cavity.[28] Several differently angled endoscopy settings are available (0˚, 30˚, 45˚, and 70˚), and these can be used to visualize the structures of the protympanum, retrotympanum, and epitympanum clearly. Secondly, high-resolution endoscopy is now available, and this can assist in distinguishing diseased mucosa from normal mucosa in the middle ear.[19] Finally, with minimal incisions and low invasiveness, endoscopic approaches are associated with reduced postoperative pain and shorter recovery times.[28] There are studies showing that endoscopic surgery for middle ear cholesteatoma result in better outcome in terms of recurrence and residual cholesteatoma,[29] also in pediatric patients.[30]

However, there are some limitations of endoscopic ear surgery. There are disadvantages due to the requirement for one-handed surgery in endoscopic approaches. Surgeons can use only one hand to manipulate the middle ear while the other holds the endoscope, and one-handed surgery can incur drawbacks in clearing the operation field.[24] If bleeding occurs, the surgeon can only use one hand to suction blood, and this can slow the procedure. Additionally, an endoscopic image does not offer three-dimensional data, and lacks anatomical depth, making endoscopic surgery challenging for inexperienced surgeons.[24] Both limitations can be overcome through training, practice, and drills of operative skill. In terms of learning curve, a recent study showed that previous surgical experience is important for gaining a learning curve in endoscopic ear surgery. The study also demonstrated that the first 5 cases are challenging, but over 30 cases make huge progression.[31] It is thought that the surgeon who had enough experience with the microscopic trans-canal approach might have little difficulty in progressing endoscopic ear surgery. Finally, an endoscope passes through the external auditory canal to reach the middle ear cavity for examination and manipulation. Sustained usage of a light bulb with strong intensity in endoscopy generates heat around the end of the endoscopic surgery, which can damage the middle ear and adjacent structures.[32] There are still benefits of using endoscope in removing cholesteatoma in middle ear regarding clinical outcomes, endoscopic approach cannot entirely replace the role of microscopic approach because of its functionally inherent differences.[33] Therefore, the surgeon needs to select suitable instruments based on their surgical experience as well as the condition of the disease.

The limitations of this study include its relatively small sample size and non-randomized setting. Also, considering the short-term follow-up of this study, these results cannot assure the superiority of the endoscopic approach over the microscopic approach for CC removal in terms of recurrence/residue or other surgical outcomes. The size of CC in endoscopic group was smaller compared to the microscopic group; therefore, further studies regarding application of endoscopy with bigger CC are necessary. The results of this study demonstrate the feasibility of the endoscopic approach for superior visualization of the entire middle ear cavity.

CONCLUSION

Endoscopic ear surgery is a newly introduced surgical technique that can effectively and safely remove CC in children. In this study, endoscopic CC removal was equal to conventional microscopic

approaches, maybe better for visualization, and it potentially showed superiority in terms of postoperative recovery, as demonstrated by a shorter hospital stay after surgery. Endoscopic approaches for removing CC are feasible and valuable methods, allowing good surgical control of CC in the hidden areas of the middle ear, especially in young patients.

Ethics Committee Approval: Ethical committee approval was received from the Institutional Review Board of Asan Medical Center (Approval No: 2019-0579).

Informed Consent: Written informed consent was obtained from all participants who participated in this study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept – J.W.C.; Design – M.Y.K.; Supervision – J.W.C.; Materials – W.S.K., J.W.C.; Data Collection and/or Processing – Y.C.; Analysis and/or Interpretation – Y.C., M.Y.K.; Literature Review – Y.C., W.S.K.; Writing – Y.C.; Critical Review – W.S.K., J.W.C.

Declaration of Interests: The authors have no conflict of interest to declare.

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