Long-term Results of Endoscopically Assisted Pediatric Cholesteatoma Surgery

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Abstract

Objective. Routine endoscopic examination during primary surgery decreased the rate of residual cholesteatoma to 18% in our early experience. Based on this, we stopped performing routine second-look surgery in children who were endoscopically free of cholesteatoma at the end of primary surgery. We sought to investigate if second-look procedures after endoscopic-assisted surgery could safely be performed only in children at a high risk of residual disease (extensive inflammation, spontaneously ruptured or fragmented cholesteatoma, residual disease intentionally left).

Study Design. Case series with chart review.

Setting. Tertiary pediatric otolaryngology practice.

Subjects and Methods. Children aged 1 to 16 years who were treated for cholesteatoma over 15-year period by a single surgeon. Extent of disease and endoscopic findings were compared with rates of residual disease. Time to diagnosis of residual disease and prognostic factors were analyzed.

Results. Forty-two children underwent endoscopically assisted middle ear surgery for cholesteatoma. Of 42 children, 7 (17%) had additional disease found by endoscopy that was missed by microscopy alone. Twelve children at high risk had second looks; 5 (42%) had residual disease. Of 30 children, 2 (7%) presented with macroscopically evident residual cholesteatoma after no planned second look on office follow-up and subsequently underwent reoperation and were cured.

Conclusions. Selective second-look surgery in high-risk children did not adversely affect outcome as compared with the low-risk group. Cholesteatoma was identified endoscopically in 7 of 42 (17%) children thought to be microscopically free of disease at initial surgery. The endoscope may aid in visualization of difficult middle ear recesses when used to complement microscopy. Further investigation with multicenter data is needed.

Keywords

cholesteatoma, chronic otitis media, endoscopy, middle ear surgery, minimally invasive surgery, pediatric otology

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Pediatric microscopic cholesteatoma surgery has a relatively high rate of recidivism, which includes both recurrent and residual disease.¹² Recurrent disease—the development of new cholesteatoma after initial eradication—is principally a function of continuing eustachian tube dysfunction rather than the thoroughness of primary surgery. Residual disease—persistence of metabolically active cholesteatoma—results from incomplete initial resection and is a target for improved methods of extirpation. Residual microscopic disease grows and becomes visible and resectable 6 to 12 months after primary resection. Since children have a high rate of residual disease after microscopic canal wall-up resection, planned second looks have been advocated to avoid further damage to the ossicles and to reduce the risk of conversion to canal wall-down anatomy.³⁴

In 1991, the senior author began routine inspection of the middle ear with endoscopes at the completion of microscopic cholesteatoma surgery. Endoscopes allow visualization of small areas of incompletely resected cholesteatoma in the recesses of the middle ear. Not surprising, at planned second look, there was a lower incidence of residual disease with this technique.⁵ Furthermore, residual disease was found principally in 3 situations: (1) where disease was intentionally left on fragile structures, (2) in severely inflamed ears where mucosal edema and blood hid disease, and (3) with widespread or spontaneously ruptured cholesteatoma where complete resection was less likely.

Based on these observations, we restricted planned second-look procedures to children with these 3 high-risk scenarios beginning in 1999. This article examines the long-term effectiveness of endoscopically assisted pediatric cholesteatoma surgery with selective second looks on the rate of residual disease. This method of endoscopic-assisted surgery is classified as observational endoscopic use, as compared with using

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the endoscope alone without otomicroscopy, which has been termed total endoscopic ear surgery.6

Methods

Record Review

After obtaining Institutional Review Board approval (Temple University Office for Human Subjects Protections protocol 21832), we queried an electronic database including 28,000 patient encounters from the senior author’s practice in the years 1999 to 2013, as listed in Microsoft Word documents. Patients were identified with the word cholesteatoma in the search function of Microsoft Word, and all results were reviewed. Children who had undergone initial cholesteatoma surgery with endoscopic assistance were included. Patients who had undergone prior surgical treatment by outside surgeons or who required a canal wall-down technique were excluded. The extent of surgery and endoscopic findings were recorded for the initial surgery and any subsequent procedure. All data were obtained through review of clinical records and operative notes.

Surgical Technique

Most cholesteatoma operations were performed with a postauricular approach. Atticotomy and/or canal wall-up mastoidectomy was included depending on the extent of disease. Following complete cholesteatoma excision under microscopic guidance, the recesses of the middle ear—including the sinus tympani, hypotympanum, eustachian tube orifice, and epitympanum—were inspected directly. A 30-degree, 2.7-mm Storz-Hopkins rod pediatric sinus endoscope (27018BA; Karl Storz, Tuttlingen, Germany) with Xenon illumination was used. Early in the series, the endoscope was viewed directly by the surgeon’s eye. With the development of 3-chip high-definition cameras and video systems, we gradually converted to off-the-monitor technique. Photodocumentation occurred in 26 of 42 cases (62%). Additional disease was removed under microscopic guidance with a McCabe elevator, whirlybird, or Goldman ball probe and the area reinspected until free of visible cholesteatoma. Ears were reconstructed with medial temporalis fascia grafts, conchal cartilage (epitympanum), and hydroxyapatite ossicular prostheses, where appropriate. Tympanostomy tubes were often placed at the time of primary surgery in children with continuing eustachian tube dysfunction.

Planned second-look procedures were performed 6 to 9 months after initial surgery. These usually included a transcanal middle ear exploration through a posterior tympanic flap. The operating microscope and 30-degree endoscope were used to identify and remove residual disease. Residual disease was defined as either known disease from primary surgery intentionally left or small keratin pearls that were not evident at initial surgery. Ossiculoplasties done at the first surgery were revisited as needed for exposure or to improve hearing. Planned third looks were performed with a similar approach only if residual disease could not be removed completely. All patients were followed with annual audiograms and serial office microscopy by the senior author.

Analysis

Data accrued included the following: patient age, length of follow-up, location of disease, presence or absence of disease in the mastoid, presence or absence of incus erosion, and intact or fragmented removal of cholesteatoma.

Results

Population

Fifty-eight children underwent surgery for congenital and secondary cholesteatomas from 1999 to 2013. Five patients had <1 year of follow-up and were excluded from analysis. Four other patients had first surgery performed by outside surgeons and were excluded as well. The endoscope was not used on an additional 7 patients. Of the 42 evaluable children, 16 were girls and 26 were boys. Mean age at diagnosis was 8.6 years (SD, 4.2 years; range, 1-16 years). Mean follow-up was 60.2 months (SD, 51.1 months; range, 12-188 months). Seven patients had congenital cholesteatoma (16.7%), and 35 had acquired cholesteatoma (83.3%). Patients were followed with regular office exams and microscopy by the senior author with annual audiograms. Of 42 children, 20 (47.6%) were followed for >5 years.

Residual Disease

Endoscopic assistance was employed during 42 first cholesteatoma operations (see Figure 1). Of the 42 patients, 7 (17%) had additional disease found by endoscopy that was missed during the microscopic phase of surgery (3 on the oval window, 2 adjacent to the pyramidal eminence, 2 in the sinus tympani). This disease was subsequently cleared during primary surgery. Twelve children (29%) had planned second-look surgery—1 for disease intentionally left on the stapes, 2 for disease on the oval window. During the scheduled second-look procedure, 2 (17%) had residual disease in locations thought to be clear on endoscopy during primary surgery.

Of the 7 patients with additional disease discovered during primary surgery by endoscopy, 4 (57%) underwent scheduled second looks, and 3 did not. Of the 4 patients who underwent a scheduled second-look procedure, 2 (50%) had residual disease. None of the 3 children observed without an operative second look developed clinically apparent residual disease with follow-up >59 months.

Of the 30 patients who did not undergo a planned second-look procedure, 2 (7%) presented with residual disease on office follow-up. Two patients with residual disease on the second look underwent a scheduled third-look procedure. These third-look patients had disease found on endoscopy during primary surgery and subsequently had residual disease during a second-look procedure. Neither patient had evidence of disease during the third look or developed demonstrable residual disease on close follow-up. Neither of the children with unexpected residual disease lost the stapes superstructure or required conversion to a canal wall-down architecture. The average time to the discovery of residual
disease from time of original diagnosis was 10.3 ± 2.8 months.

Discussion

Cholesteatoma surgery begins with the standard goals of eradicating disease, preserving and/or restoring of hearing, maintaining temporal bone anatomy, and preventing recurrence. Complete resection or exteriorization of cholesteatoma is paramount among these. The middle ear and its recesses are frequent sites of incomplete removal during microscopic surgery. The “straight-ahead” view of the operating microscope provides limited exposure. The Buckingham mirror was invented to improve vision but proved ineffective. Atticotomy and transmastoid approaches to the facial recess, sinus tympani, and hypotympanum provide access but are time-consuming and remove important barriers to recurrent retraction if eustachian tube dysfunction continues.

The introduction of small, high-quality, angled rigid endoscopes has improved visualization of the middle ear recesses not well seen with the operating microscope. Endoscopically assisted surgery and, more recently, transcanal entirely endoscopic procedures have been advocated in the management of middle ear cholesteatoma in adults. The newest generation of slim endoscopes and high-definition cameras, as well as dedicated instrument sets, makes these approaches possible.

In a series of adult cholesteatoma operations, Thomassin et al compared endoscope-aided procedures with traditional microscopic surgery and found reduction in residual cholesteatoma from 47% to 6% during second-look procedures. In a more recent series of adult patients (169 patients, 184 ears) undergoing endoscopically assisted adult cholesteatoma surgery, the overall recurrence rate was 24 of 184 (13%). The unexpected residual rate was 5 of 184 (3%). The use of endoscopes allowed for the avoidance of a mastoidectomy in 65 (35%) of 184 ears.

The literature describing pediatric middle ear endoscopy and its application to congenital and acquired cholesteatoma remains scant. Rosenberg et al and Good and Isaacson reported early series of endoscopically assisted operations with good outcomes. James described good results during a 10-year experience with an endoscopically assisted approach and his gradual migration to endoscopic transcanal surgery in selected children.

Recidivism in cholesteatoma surgery—encompassing both residual and recurrent disease—is more common in the pediatric population. Aggressive disease becomes symptomatic at an early age. Similarly, young children are likely to have continuing eustachian tube dysfunction, placing them at risk for retraction pocket formation and recurrent cholesteatoma after successful initial surgery and reconstruction. Endoscopic assistance can reduce the risk of recurrence. Re-retraction is less likely if healthy native tympanic membrane or portions of the scutum can be preserved as barriers. Still, the principal benefit of endoscopy is completeness of cholesteatoma excision—that is, reducing the incidence of residual disease. Endoscopic guidance achieves this. At the completion of the microscopic phase of surgery, 7 of 42 children in our study had additional disease that was identified on endoscopic examination.

Of the 7 children with disease found by endoscopy but not microscopy at first operation, 4 met our criteria for planned second looks. At those planned second looks, 2 of 4 children (50%) had residual disease. This demonstrates the power of the endoscope in identifying disease not otherwise visible on microscopy. It also attests to the value of planned second-look surgery in high-risk situations, even in endoscopically clean ears.
Enhanced detection and immediate removal of microscopically occult disease potentially reduce the need for planned second-look procedures. In our initial series of endoscopically assisted pediatric cholesteatoma surgery, the rate of residual disease during routine second-look procedures was relatively low (18%). Thus, 82% of second operations might be avoided with proper selection. We therefore decided to restrict planned second-look procedures to those children with high risk of residual disease. These included those with inadequate visualization due to inflammation, residual disease intentionally left on sensitive structure, or extensive disease or spontaneously ruptured cholesteatoma for which complete eradication was less certain.

Based on this selective second-look approach, our rate of unexpected residual disease overall was 4 of 42 patients (9.5%). This compares favorably with adult series (3%-6%) based on endoscopically assisted approaches for cholesteatoma removal. The selective second-look protocol correctly predicted 5 of the 7 cases of residual cholesteatoma. The 2 unexpected incidences of residual cholesteatoma were identified early by office examination (mean time to discovery, 12.5 months). In neither case was the stapes superstructure destroyed by disease, nor was conversion to canal wall-down configuration required.

Planned second-look procedures in children with high-risk features resulted in finding residual cholesteatoma in 2 of 12 (17%) patients versus 2 of 30 (7%), P = .56, Fisher’s exact test. Statistical significance could not be achieved, possibly due to the need for more subjects. Of the 12 (33%) patients judged to be at high risk for residual disease, 4 (33%) had additional cholesteatoma identified by endoscopy, as compared with 3 of 30 (10%, P = .09) low-risk patients. The use of the endoscope potentially prevented residual disease in 3 of 42 (7%) patients.

There is much variability in the reported incidence of residual cholesteatoma. Several factors play a role in this, including surgeon experience and skill level, time until revision surgery for residual cholesteatoma, and extent of primary surgery. In our series, residual disease was discovered at a mean of 10.3 months. Although follow-up is important, particularly for monitoring for recurrence, our series suggests that residual disease is unlikely to present more than a year after surgery when endoscopic assistance is used to ensure adequate disease removal. The 2 patients with residual disease who did not undergo a planned second-look procedure presented early as well (average of 12.5 months). Delayed presentation of residual disease in those patients who did not undergo a planned second-look procedure did not result in a more extensive salvage operation.

It is our opinion that endoscopy is useful in all pediatric cholesteatoma cases with disease medial to the ossicles. Visualization of the sinus tympani and hypotympanum is improved as compared with that of microscopy. Small disease in and around the stapes footplate can be eradicated with greater assurance with the addition of endoscopy, as improved visualization is key to eradicating disease.

This study is limited by its retrospective nature and relatively small population. Early operations in the series were performed by looking directly through the endoscope, with gradual progression to more optimal 3-chip high-definition camera and video systems. However, 2 cases of residual cholesteatoma occurred within the first 7 years of the study, and the remaining 5 cases occurred over the last 8 years of the study. It is possible that there may be clinically undetectable residual disease in some of the patients being actively followed in the cohort. The endoscope may have missed microscopic disease, which possibly explains the 2 patients of the 30 who did not have a second-look procedure who subsequently had residual disease. In children judged to be at low risk of residual disease, endoscopic evaluation identified residual cholesteatoma in 3 of 30 (10%) patients during primary surgery but may have missed residual disease in 2 of 30 (7%) patients. Because of this, close follow-up is necessary, as there are still instances of unexpected residual disease even with improved endoscopic visualization. Furthermore, there is a learning curve in difficult operations. The chance of favorable results improves with a surgeon’s experience, introducing a potential bias over time. These factors limit the generalizability of our results to other surgical settings. Further investigation is needed.

Conclusion
The addition of endoscopic assistance during pediatric cholesteatoma surgery has been shown to limit the extent of dissection and can aid in detecting additional disease that may otherwise be missed with otomicroscopy. While variable rates of residual cholesteatomas are reported with the use of endoscopes, the positive effect of improved visualization is widely accepted. Our data support the effectiveness of endoscopic guidance in ensuring complete cholesteatoma removal, especially from the middle ear recesses. Selective second-look surgery is a reasonable choice in children deemed endoscopically free of disease at the completion of primary surgery. We did not demonstrate a difference in residual rate between selective endoscopic groups deemed high and low risk. While continued follow-up is needed to watch for recurrence, clinically evident residual disease presented at an average of 12.5 months in this series. Otologists may be selective in recommending second-look procedures for pediatric cholesteatoma that is well removed at first surgery, as there was no difference demonstrated in those who underwent selective second look for high-risk disease and those who did not.

Author Contributions
David Sarcu, acquisition and analysis of data, drafting and revising manuscript; Glenn Isaacson, design of study, analysis of data, drafting and revising manuscript, final approval.

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