

## Case Report

# Complete Removal of Stage II Congenital Cholesteatoma Destroying Temporomandibular Joints Using Transcanal Endoscopic Ear Surgery With a Mirror Technique: A Case Report

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Transcanal endoscopic ear surgery (TEES) provides wide-angle clear vision for otologic surgery. We report the utility of TEES with the mirror technique for the complete removal of a congenital cholesteatoma in a 3-year-old boy. A white mass was observed through the tympanic membrane, and a congenital cholesteatoma was suspected during the conservative treatment of otitis media with effusion. Pre-operative computed tomography (CT) revealed an irregular mass lesion in the left middle ear, with bone erosion in the hypotympanum. During surgery, an open-type cholesteatoma was observed, mainly in the middle lower tympanum. The cholesteatoma had destroyed the periphery of the temporomandibular joint, which was widely exposed. The cholesteatoma had also spread to the tympanic sinus. Careful observation with a forward-oblique viewing endoscope and a variable angle tympanic mirror enabled complete removal of the mass under endoscopic guidance. No recurrence was observed during the postoperative follow-up, although residual open-type congenital cholesteatoma may often result in recurrence. We believe that careful removal of the lesion under TEES, which allowed us to perform surgery under a secure view with illumination and magnification, may have facilitated complete removal and prevented recurrence.

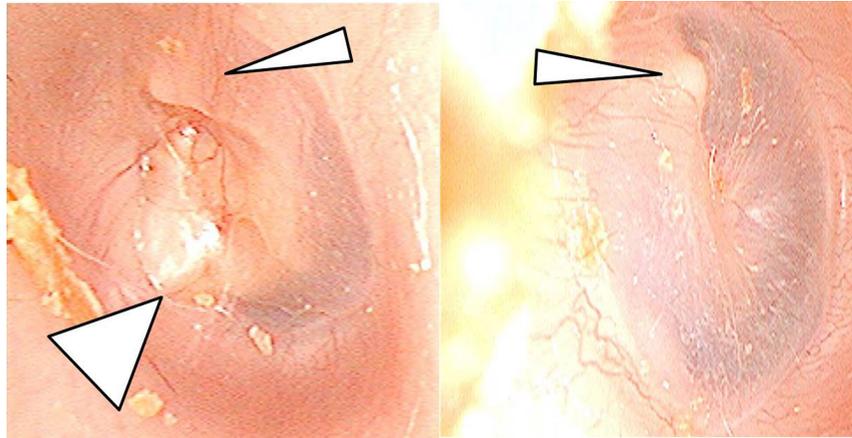
**KEYWORDS:** Congenital cholesteatoma, tympanoplasty, transcanal endoscopic ear surgery

## INTRODUCTION

Transcanal endoscopic ear surgery (TEES) has gained popularity as a new middle ear surgical technique. Middle ear surgery with TEES requires a comparatively smaller skin incision and less ear canal bone removal than with conventional microscopic surgery. The advantages of endoscopy include better illumination and a wide-angle field of view of even the deep parts, thereby reducing the blind spot.<sup>1</sup> An incision in the ear canal, or a postauricular incision is necessary to ensure a good surgical field for middle ear surgery under microscopic guidance. However, parts of the tympanic cavity and the anterior epitympanic recess still lie in an anatomical blind field. In this study, we present a case of congenital cholesteatoma treated using TEES to prevent residual lesions, because the lesions were located in the deep lower tympanic cavity. The extensive congenital cholesteatoma involving the tympanic sinus, hypotympanum, and temporomandibular joint capsule was resected completely using TEES combined with the mirror technique.

## CASE PRESENTATION

The patient was a 3-year-old boy who was referred to our otolaryngology department by a physician for a suspected left congenital cholesteatoma. During his first visit, a white mass was observed behind the left tympanic membrane (Figure 1). The right tympanic membrane was normal. Temporal bone computed tomography (CT) revealed a mass in the left middle ear (Figure 2A and B). The mass, which resembled a congenital cholesteatoma, had progressed toward the hypotympanic space and temporomandibular joint. Partial bone destruction was observed in the temporomandibular joint. There was no progression toward the superior tympanic cavity, tegmental wall, or mastoid air cells. We diagnosed the condition as stage II pars tensa cholesteatoma involving the tympanic cavity (T) and sinus tympani (S2).<sup>2</sup> We decided to perform TEES because the mass extended from the lower-anterior part of the tympanic



**Figure 1.** Tympanic findings (a) Left tympanum on the affected side; a white mass was seen through the left tympanum; (b). The right tympanum was normal. (thin arrow: malleus, thick arrow: cholesteatoma).

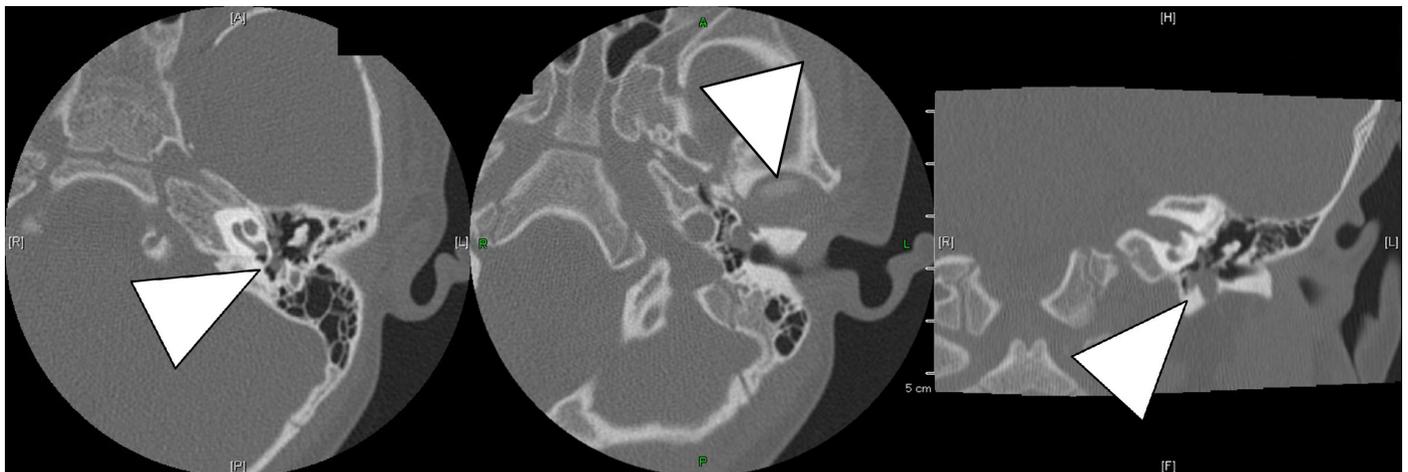
cavity, which could be easily accessed with the endoscopic approach. Moreover, in this case, the space for inserting the endoscope was safe because the outer ear canal was fairly straight and not stenotic.

Type I Tympanoplasty was performed with TEES under general anesthesia. We observed an open-type cholesteatoma extending from the anterior to the deep part of the tympanic cavity after elevating the tympanomeatal flap. The chorda tympani was identified and preserved. The incudostapedial joint was dislocated. The superior tympanic cavity was intact, without extension of the cholesteatoma. The cholesteatoma had progressed into the lower tympanic cavity, as observed under clear vision with the angled endoscope. The lower part of the external ear canal was drilled to expose the hypotympanum, which would allow surgical manipulation under a straight endoscope. Subsequently, the bottom of the lesion was visualized, where the temporomandibular joint capsule was exposed by osteolysis caused by the cholesteatoma (Figure 3).

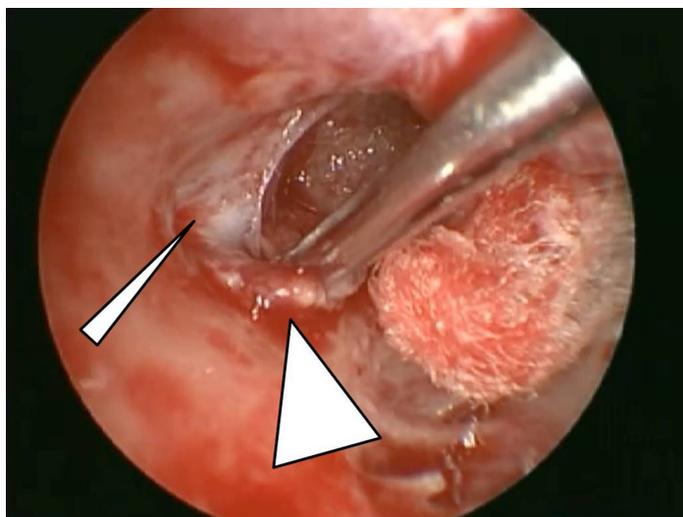
The area occupied by the cholesteatoma extended from the anterior tympanum, right beneath the tensor tympani tendon, onto the joint capsule, where the cholesteatoma matrix was carefully

elevated and released. This enabled closer observation under the endoscope, deep into the tympanic sinus: the matrix became fully visible using a 0° endoscope combined with an adjustable angle tympanic mirror (Figure. 4). We confirmed the presence of remnants of the matrix within the tympanic sinus through the 0° endoscope and tympanic mirror, which were removed under direct vision under 30° endoscopy by shaving the bone (since we could determine the exact location of the remnants). The matrix was completely removed by alternating indirect observation under the tympanic mirror and the 0° endoscope, with manipulation under the direct view of the 30° endoscope. Finally, surgery was completed under careful observation of the whole area, including the tympanic sinus, using the straight endoscope and the tympanic mirror.

Postoperative complications, including vertigo/dizziness and facial palsy, were not observed. The external auditory canal was completely epithelized. Perforation of the tympanic membrane was not observed during 6 months of follow-up. CT performed 1 year postoperatively did not reveal evidence of recurrence. The peep show test was performed one and a half years later; there was no difference in hearing between the 2 sides (Figure 5). The patient is still being followed up.



**Figure 2.** Pre-operative temporal bone computed tomography (CT). (A) (Left): Temporal CT (axial) showing a mass in the tympanic sinus (arrowhead). There was no abnormality in the ossicular chain. (B) (Center): Exposure of the temporomandibular joint by the cholesteatoma. (arrowhead: mandibular condyle) (C) (Right): Temporal bone CT (coronal) showing progression of the mass to the bottom of the hypotympanum (arrowhead) and destruction of its floor, with exposure of the temporomandibular joint capsule.



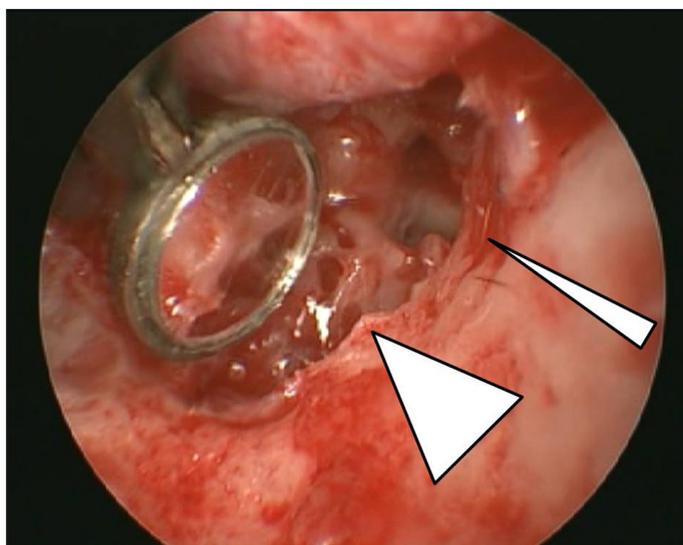
**Figure 3.** Temporomandibular joint capsule and matrix (thin arrow, temporomandibular joint capsule; thick arrow, cholesteatoma matrix).

Written informed consent were obtained from the patient who participated in this study.

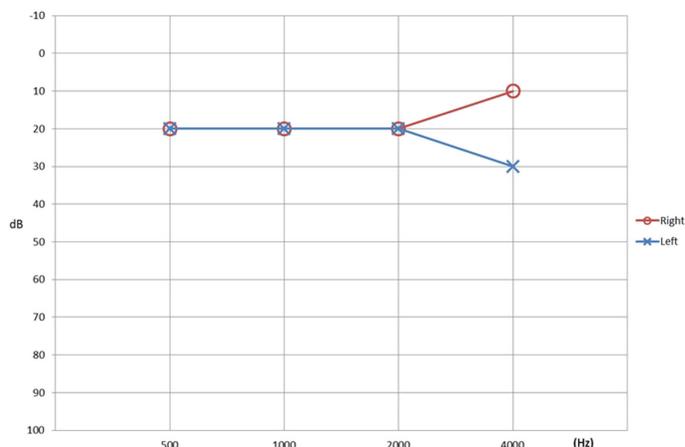
**DISCUSSION**

TEES has enabled in-depth visualization of the narrower areas of the ear canal with better clarity and illumination. Reports on the indications of TEES for treating childhood congenital cholesteatoma have increased recently.<sup>3</sup>

The recurrence of congenital cholesteatoma is higher than that of acquired cholesteatomas. Nelson et al<sup>4</sup> reported a recurrence rate of 34% (33/98) in a meta-analysis of microscopic surgery for 119 patients aged 2-14 years in the United States. Residual recurrence was observed in about 10-50% of cases during the second surgery in staged tympanoplasty.<sup>5</sup> The sinus tympani, the area around the stapes, and others were most commonly associated with remnants of cholesteatoma, which are anatomical blind spots with microscopy.



**Figure 4.** Observation of the bottom of the tympanic sinus with the tympanic mirror. The cholesteatoma matrix beneath the deep tympanic sinus was visualized in the mirror. The matrix was removed after the observation. (thin arrow: chorda tympani, thick arrow: edge of the tympanic sinus).

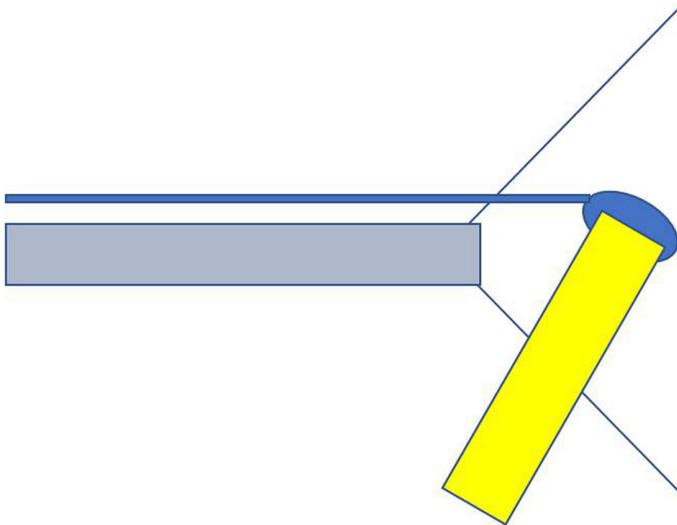


**Figure 5.** Peep show test (circle mark: right ear, cross mark: left ear). There was no difference in hearing between the 2 sides.

Owing to the higher recurrence rates, TEES has now become popular for congenital cholesteatoma, because observation and manipulation under the endoscope may reduce the likelihood of remnants.<sup>6</sup> Thomassin<sup>7</sup> first reported in 1993 that microscopic surgery combined with the endoscope reduced the persistence and recurrence of cholesteatomas. Remnants and recurrence were observed in 21 (47.7%) of the 44 cases treated using only microscopic surgery between 1979 and 1985, whereas relapse was observed in only 2 of 36 cases treated with endoscopy-assisted surgery between 1985 and 1991.<sup>7</sup> Kozin and David<sup>6</sup> reported that endoscopy performed after microscopic surgery detected remnants in 8.6-50% of cases. Few studies have performed a direct comparison of microscopic and endoscopic surgery. Ghadersohi et al<sup>8</sup> reported that there were no statistical differences in the residual rate, recurrence rate, or complications with both methods. Park et al<sup>9</sup> reported that recurrence was observed in only 1 of 25 patients who underwent TEES for congenital cholesteatoma after a 2-year follow-up. Hence, TEES can ensure proper resection and control of congenital cholesteatomas. Since the posterior ear incision is not required, which is an advantage of TEES, this approach is particularly suitable for children with respect to post-operative management.

This case of congenital cholesteatoma was equivalent to stage II cholesteatoma, with deep invasion into the tympanic cavity. We selected TEES for better visualization of the lesion.

Otologic surgery, especially ensuring complete remission of cholesteatomas, is most challenging in the tympanic sinus. Deep tympanic sinuses are sometimes very difficult to visualize even with oblique viewing endoscopes. Marchioni et al<sup>10</sup> classified the tympanic sinus into types A, B, and C depending on the positional relationship of its depth with the facial nerve. The deepest part of type A tympanic sinuses can be observed only under a microscope. Our patient had a type C tympanic sinus (Figure 2A). Complete removal of the cholesteatoma seemed relatively difficult because the sinus was deep and the matrix had spread (open-type cholesteatoma). Therefore, we used a tympanic mirror. A 3.2-mm mirror was attached to the tip of the ear forceps, whose angle could be changed from 0° to 50°. This technique has been used with microscopic surgery.<sup>11</sup> Using a mirror with the TEES reduced the anatomical blind spots, which allowed us to observe all the remnants of the pathological cholesteatoma



**Figure 6.** Schematic illustration of the observation with a tympanic mirror under TEES. This technique enabled us to observe blind spots, such as that indicated by the asterisk. TEES, Transcanal endoscopic ear surgery.

epithelium. The wide-angle lens of the endoscopes used in TEES provided a wide view of 70°. Therefore, a 0° endoscope coupled with a 50° tympanic mirror can provide a 100° view (Figure 6). Thus, we successfully investigated the bottom of the tympanic sinus (denoted by the asterisk in Figure 6), resulting in surgical success and complete remission.

## CONCLUSION

We achieved better visualization of our patient's cholesteatoma, which had progressed to the deep part of the tympanic sinus, using an endoscope combined with an adjustable mirror. The cholesteatoma matrix, which had eroded the mandibular joint, could be removed with closer observation using endoscopy after canalplasty. Therefore, TEES is useful because it is minimally invasive and allows careful observation.

**Ethics Committee Approval:** This study was conducted in accordance with the approval of the ethics committee of the Keio University School of Medicine (approval no. 20200033).

**Informed Consent:** Written informed consent were obtained from the patient who participated in this study.

**Peer Review:** Externally peer-reviewed.

**Author Contributions:** Concept - S.O., M.H., M.F.; Design - S.O., M.H., M.F.; Supervision K.O.; Resource - S.O., M.H., M.F.; Materials - S.O., M.H., M.F.; Data Collection and/or Processing - S.O., M.H., M.F.; Analysis and/or Interpretation - S.O., M.H., M.F.; Literature Search - S.O., M.H.; Writing - S.O., M.H.; Critical Reviews - K.O.

**Conflict of Interest:** The authors have no conflict of interest to declare.

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