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Case Report

A Rare Case of Bifurcated Chorda Tympani

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Preservation of the chorda tympani is important in middle ear surgery to prevent dysgeusia postoperatively. However, determining the exact course of the chorda tympani before surgery is not always possible, especially in cases with accompanying malformations. In this report, we presented an extremely rare case of bifurcation of the chorda tympani in a 15-year-old male patient. We performed tympanoplasty for a middle ear malformation with conductive hearing loss. During the operation, we noticed and carefully preserved the bifurcated chorda tympani. The patient did not develop dysgeusia postoperatively. Appropriate handling and understanding of the anomalous chorda tympani preserved the patient's sense of taste and hence quality of life.

KEYWORDS: Chorda tympani, middle ear malformation, hearing loss, tympanoplasty

INTRODUCTION

The chorda tympani is a branch of the facial nerve that separates off above the stylomastoid foramen, runs through the canaliculus, and then appears on the lateral wall of the tympanum. Thereafter, it passes forward between the malleus and incus through the petrotympanic fissure, emerges in the temporal fossa, joins the lingual nerve, and finally reaches the floor of the mouth. The chorda tympani is responsible for the sensation of taste in the anterior two-thirds of the tongue and the secretion of saliva from the submandibular and sublingual glands. The chorda tympani can be seen during tympanoplasty when creating the tympanomeatal flap, and its preservation is important for preventing dysgeusia postoperatively.

Malformation of the chorda tympani usually occurs as part of a facial nerve anomaly and is often accompanied by malformation of the external ear or ossicular chain [1]. Malformation of the facial nerve in isolation is rare, and cases involving only the chorda tympani are even rarer [1]. Two cases of duplication of the facial nerve were identified in 972 patients undergoing cochlear implantation [2]. However, malformation of the facial nerve is usually associated with malformation of the external ear; in one report, this association was found in approximately 65% of cases [3].

Few reports on the bifurcation of the main trunk of the facial nerve between the inner ear canal and mastoid segment are available [4-7], but reports on the bifurcation of the chorda tympani are extremely rare [8]. In the current report, we present a very rare case of bifurcated chorda tympani accompanied by malformation of the external ear.

CASE PRESENTATION

A 15-year-old male patient with 22q11.2 deletion syndrome, primary hypoparathyroidism, and ventricular septal defect presented to our department with conductive hearing loss in the right ear. At the age of three, he had undergone bilateral tympanic tube insertion for chronic otitis media with effusion. At the age of eight, he had undergone bilateral myringoplasty to repair bilateral perforation of the tympanic membrane that had occurred five years earlier at the time of tube insertion. His hearing had subsequently improved on the left side but not on the right.

His right external auditory canal was narrow and the right tympanic membrane was small (Figure 1). An audiogram showed conductive hearing loss on the right (Figure 2). The long process of the incus could not be visualized clearly on a computed tomography scan (Figure 3). The course of the facial nerve appeared to be normal. We suspected ossicular malformation to be the cause of his conductive hearing loss and that tympanoplasty would improve his hearing. The patient and his family agreed to the surgery.

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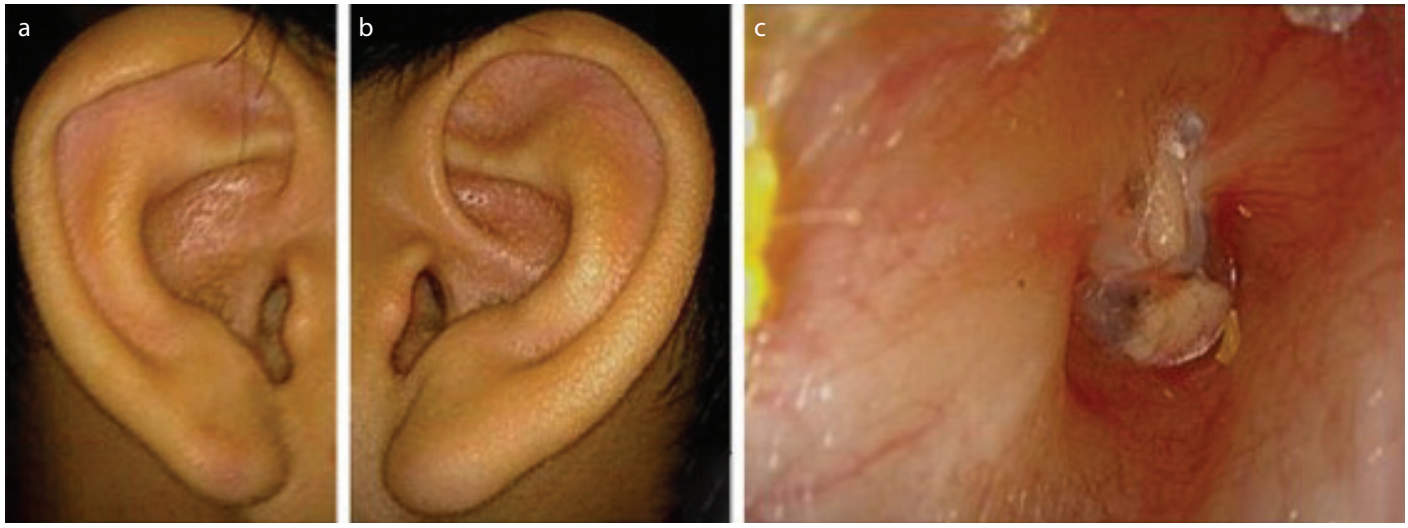

Figure 1. a-c. Clinical photograph showing that the right auricle has a mild malformation compared with the left auricle (a, b). The right external auditory canal is narrow and the right tympanic membrane is small (c).

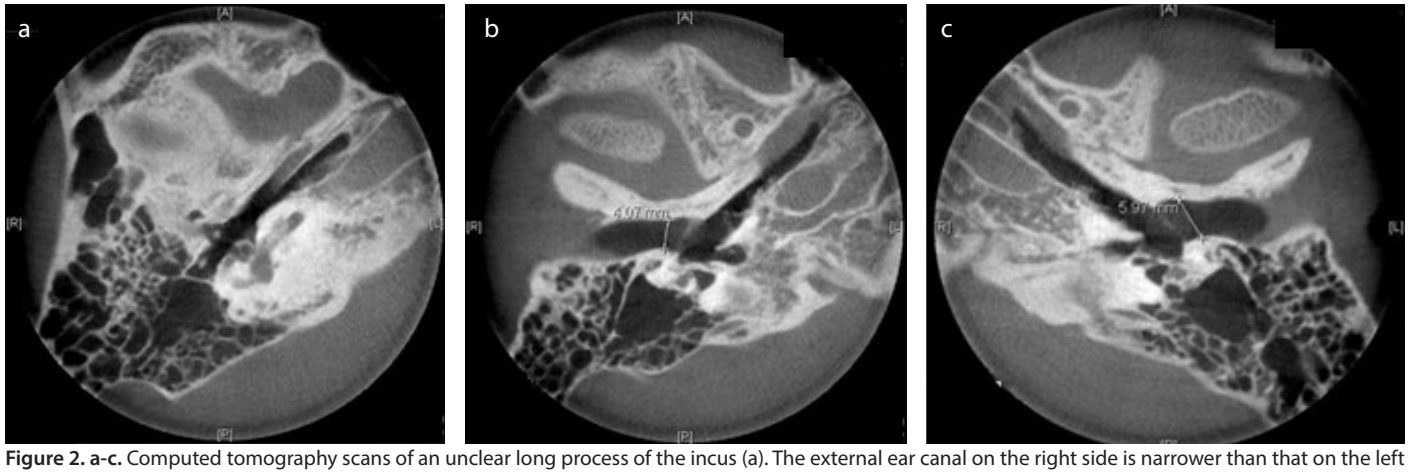

Figure 2. a-c. Computed tomography scans of an unclear long process of the incus (a). The external ear canal on the right side is narrower than that on the left side (b, c).

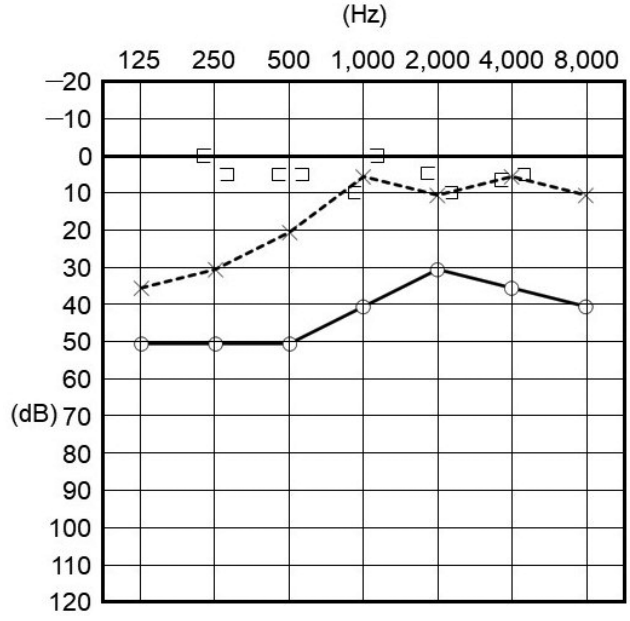

Figure 3. Preoperative audiogram of conductive hearing loss in the right ear.

Under general anesthesia, the tympanomeatal flap was elevated to access the tympanic cavity. We confirmed the ossicular chain malformation at this point. The neck of the malleus was located vertically and caudally accompanying its shortened head. The long process of the incus was shortened and located caudally. The footplate of the stapes, which had good mobility, was rotated 60° in a clockwise direction and was slightly thick, and the head of the stapes was hypoplastic. The incudostapedial joint was replaced with a membrane-like structure; after removal of this structure, we recognized the gap between the long crus and the head of the stapes.

While scraping the posterior wall of the tympanic cavity to create the working space for tympanoplasty, we found that the chorda tympani was bifurcated within the bone, with one branch running normally between the malleus and incus and the other running toward the lower anterior aspect of the tympanum in the direction of the petrotympanic fissure (Figure 4). Although both branches were thought to be running into the petrotympanic fissure, their entry to the fissure could not be confirmed due to the limited anterior surgical field. We suspected an accompanying facial nerve malformation but found that the facial nerve was normally running on the head side of the stapes with no malformation. Preserving the bifurcated

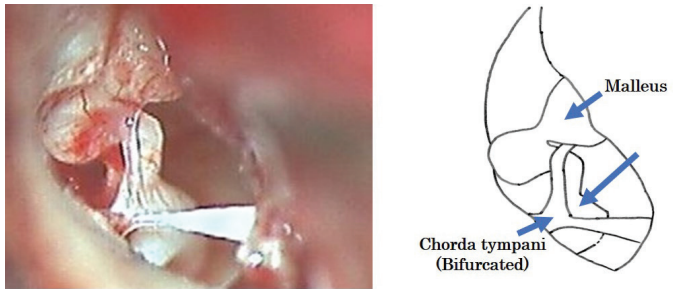

Figure 4. Intraoperative photograph of the bifurcation of the chorda tympani, with one branch running between the malleus and incus and the other running toward the lower anterior aspect of the tympanum.

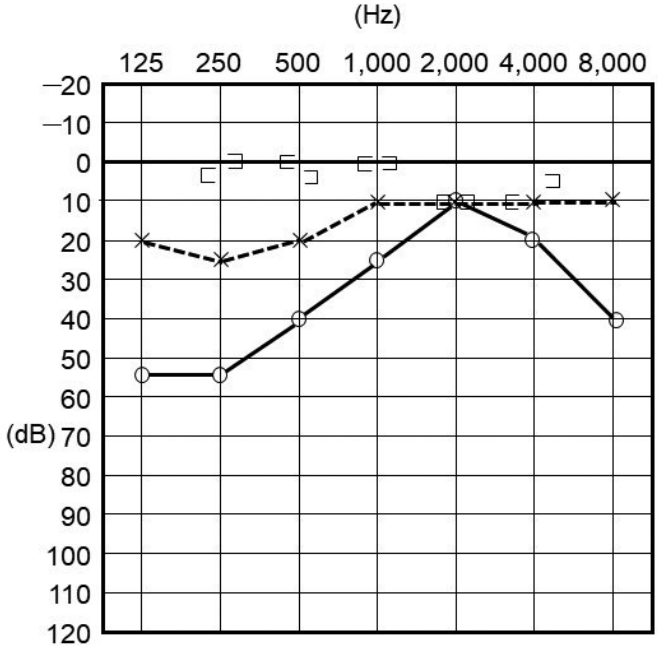

Figure 5. Postoperative audiogram confirming hearing improvement.

chorda tympani successfully, we completed the scraping of the posterior wall.

We collected and inserted the necessary auricular cartilage into the gap between the incus and stapes. After confirming improved conduction, we fixed the reconstructed ossicular chains with fibrin glue.

The patient had no taste disorder, dizziness, or nystagmus postoperatively and was discharged on the second postoperative day. Pure tone audiometry performed four months after surgery confirmed improved hearing on the right side (Figure 5).

The case patient provided written informed consent.

DISCUSSION

This extremely rare malformation of the chorda tympani is believed to be related to developmental abnormalities of the ossicular chains because most of the reported cases have had accompanying congenital ossicular chain malformation. In a study of congenital ear deformities, the incidence rates of facial nerve anomalies were 45.2% and 60.8% in patients with aural atresia and middle ear deformities, respectively [9]. The malleus and incus develop from the first branchial arch, and the stapes develops from the second branchial arch. The external ear canal and eardrum develop from the first branchial arch and the facial nerve develops from the second branchial arch. Our patient had a small external ear, narrowing of the ear canal and eardrum, and ossicular malformation affecting all three ossicular bones. Although scholars have argued that the pathogenesis of these malformations could not be simply explained by a branchial-based theory [10], we believe that the abnormalities in our patient occurred during development of the first and second branchial arches.

Several types of ossicular malformation have been reported. Facial nerve anomalies are common in patients with external auditory canal atresia or dysplastic inner ears; by contrast, facial nerve anomalies associated with congenital footplate fixation in the presence of normal external and inner ears are rare[6]. Several abnormalities of the facial nerve have been reported, including gross overhang of the naked nerve, bifurcation of the nerve around the oval window, the nerve running on the promontory between the oval and round windows, and duplication (not bifurcation) of the chorda tympani[8]. Only four ears (from three patients) with bifurcated chorda tympani have been reported. In three of these cases, the bifurcations traveled together between the malleus and incus in a normally running root; in the fourth case[8], after bifurcation, one branch traveled normally between the malleus and incus and the other traveled between the malleus and tympanic membrane. Our present case is only the second in which one of the bifurcations of the chorda tympani did not travel between the malleus and incus.

CONCLUSION

Iatrogenic nerve injury leading to taste disorder can occur when tympanoplasty is performed without knowledge of the potential ossicular, auricular, or external ear canal malformation of the chorda tympani in patients. Although several reports are available on the facial nerve traveling abnormally, bifurcation of the chorda tympani is extremely rare. Facial nerve abnormalities originate from the abnormal development of the branchial arch, and many cases are accompanied by outer and middle ear malformation. Care is required intraoperatively to avoid damage to the chorda tympani, especially in patients with accompanying malformations.

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

Peer-review: Externally peer-reviewed.

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

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