

CASE REPORT

Extensive Glomus Tympanicum Mimicking Chronic Granulomatous Otitis Media

Kai-Chieh Chan, Shiang-Fu Huang, Jen-Liang Su, Che-Ming Wu

Department of Otolaryngology, Chang Gung Memorial Hospital, Chang Gung University, Taipei, Taiwan

Glomus tympanicum, a hypervascular tumor in the middle ear, is usually suspected based on pulsatile tinnitus and retrotympanic reddish mass on otoscopic examination. These hallmarks may be masked by other middle ear diseases. Herein, we report an unusual case of the extensive glomus tympanicum masqueraded as chronic granulomatous otitis media, which was extirpated without preoperative embolization via canal wall-down mastoidectomy approach with piecemeal adrenaline-soaked cotton pledgets wrapping and compression under controlled hypotension intraoperatively, producing minimal blood loss. There was no significant intraoperative complication and no evidence of recurrence over 1 year of follow-up.

Submitted : 26 June 2008

Revised : 19 August 2008

Accepted : 24 December 2008

Glomus tumors (paragangliomas), which are more commonly found in Caucasians than in Orientals, originate from neuroendocrine cells of the extra-adrenal paraganglia of the autonomic nervous system. Paragangliomas of the head and neck occur with an approximate incidence of 1 in 30,000 and carotid body tumors are the most common, followed by jugulotympanic and vagale tumors. In the temporal bone, the glomus jugulare is the most common, followed by the glomus tympanicum (GT)^[1,2].

Surgical excision is the mainstay of treatment for the GT. Intraoperative massive bleeding often bothers surgeons while removing tumors. Therefore, preoperative angiography and embolization were advised in some cases^[1-3]. In this article, we will discuss the dilemma and surgical technique of how to manage an extensive middle ear hypervascular tumor without preoperative embolization, of which the diagnosis from frozen section is inconsistent with that from permanent section-paraganglioma.

Case Report

A 29-year-old Taiwanese female visited our clinic complaining of a three-month history of left hearing

impairment and occasional otorrhea. Tinnitus, dizziness, and facial paralysis were unremarkable. Otoscopic examination revealed a reddish polypoid mass protruding through the left tympanic membrane (Figure 1). The pure tone audiometry in the left ear was 43dB, and air-bone gap was 38 dB (Figure 2a). The high resolution computed tomography (CT) of temporal bone demonstrated that a low-density mass with ossicular involvement occupied the entire tympanic cavity extending to the external ear canal and mastoid antrum (Figure 3). On suspicion of left chronic granulomatous otitis media, a tympanomastoidectomy was performed. During the operation, the mass was partially resected and then was sent for frozen section examination because of easy-touch bleeding. The initial frozen section was reported as "inflammatory granulation tissue". Then the surgical procedure was proceeded, and the tympanomastoid cavity was exposed via posterior tympanotomy with a post-auricular approach. Besides, the systolic blood pressure and mean arterial pressure of the patient were regulated to a controlled hypotensive status perioperatively (80-90 and 50-65 mm Hg, respectively). The tumor was found to occupy the

Corresponding address:

Dr. Che-Ming Wu

Department of Otolaryngology Chang Gung Memorial Hospital

5, Fu-Shin Street, Kweishan Taoyuan 333, Taiwan

Phone: 886-3-3281200 ext.3967; Fax: 886-3-3979361; E-mail: bobwu506@seed.net.tw

Copyright 2005 © The Mediterranean Society of Otolaryngology and Audiology

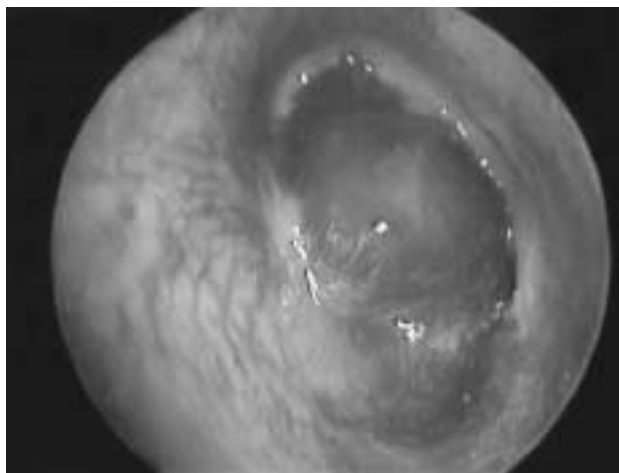


Figure 1. Otoscopy shows a reddish granulomatous tumor protruding through the tympanic membrane with some whitish discharge coating.

holotympanic cavity with extension to antrum. The malleus and incus were so tightly encased by the mass as to make their free dissection and preservation arduous. The stapes and facial canal were abutted by the tumor without invasion. A canal wall-down approach was performed so that the tumor could be wrapped and compressed completely by piecemeal 1:100,000 adrenaline-soaked cotton pledgets (Figure 4a). Then the tumor shrunk gradually and the

supplying vessels of the tumor could be identified easily. Therefore, the entire tumor along with the malleus and incus could be excised with the aid of bipolar diathermy (Figure 4b), and the type III tympanoplasty and meatoplasty were carried out eventually. The operative blood loss was only 120 ml and intraoperative complications were absent. Grossly, the tumor was measured about 15x12x10 mm; microscopically, the uniform tumor cells were arranged in the Zellballen formation. The pathological diagnosis demonstrated a “paraganglioma”. The postoperative course was uneventful, and the following magnetic resonance imaging (MRI) revealed no glomus tumor in the jugular bulb or carotid artery. Therefore, the final diagnosis of “Fisch type B glomus tympanicum” was confirmed. The patient’s hearing threshold improved to 33 dB with a 20 dB air-bone gap (Figure 2b), and there was no evidence of recurrence over 1 year of follow-up.

Discussion

GT is a highly vascular, slow-growing neoplasm that arises from the glomus body and runs with the tympanic branch of the glossopharyngeal nerve (Jacobson’s nerve).

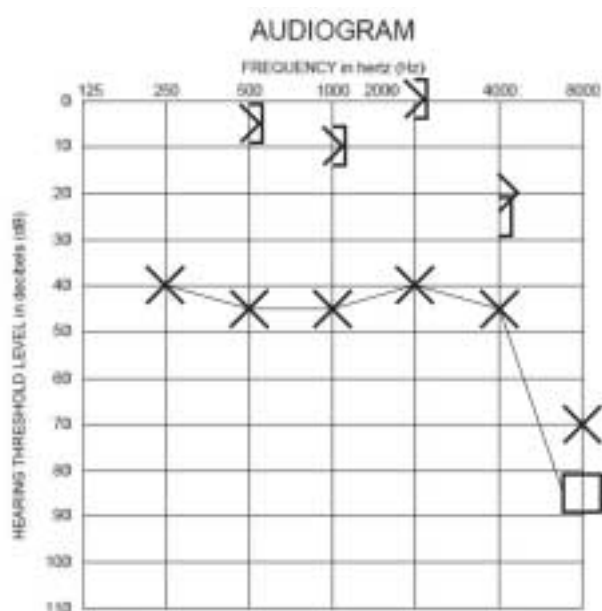


Figure 2a

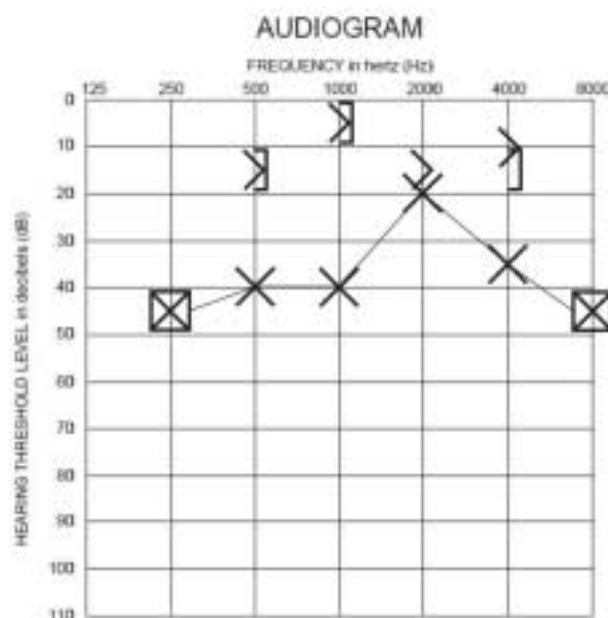


Figure 2b

Figure 2. (a) Audiogram shows the preoperative hearing threshold (Oct., 2007) (b) Audiogram shows the postoperative hearing threshold (Nov., 2008)

The diagnosis of GT usually depends on the clinical symptoms and evidences from the otoscopy and imaging studies. Pulsatile tinnitus is the most common presenting symptom, followed by conductive hearing loss^[1,2]. Upon otoscopy, GT is typically recognized as retrotympanic reddish tumor behind the intact eardrum. In our case, however, a protruding middle ear granulomatous mass presenting with unilateral hearing loss and otorrhea without pulsatile tinnitus might lessen the initial suspicion of GT. Besides, on CT scan, GT classically

reveals as a round soft-tissue density on the cochlear promontory, limited medially to the tympanic membrane, and often is not associated with any ossicular erosion. Nevertheless, our case showed a holotympanic soft tissue density mass with ossicular encasement and mastoid antrum involvement on the CT. Therefore, it could hamper the differential diagnosis of GT from chronic granulomatous otitis media preoperatively.

The preoperative angiography and embolization are often advocated before resecting glomus tumors, but

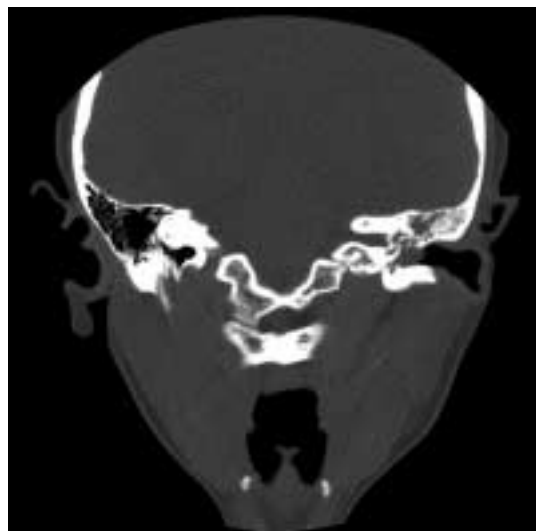


Figure 3. CT, coronal view: a soft tissue density mass occupying the left holotympanic cavity with extension to antrum and protruding through the tympanic membrane to the external auditory canal.

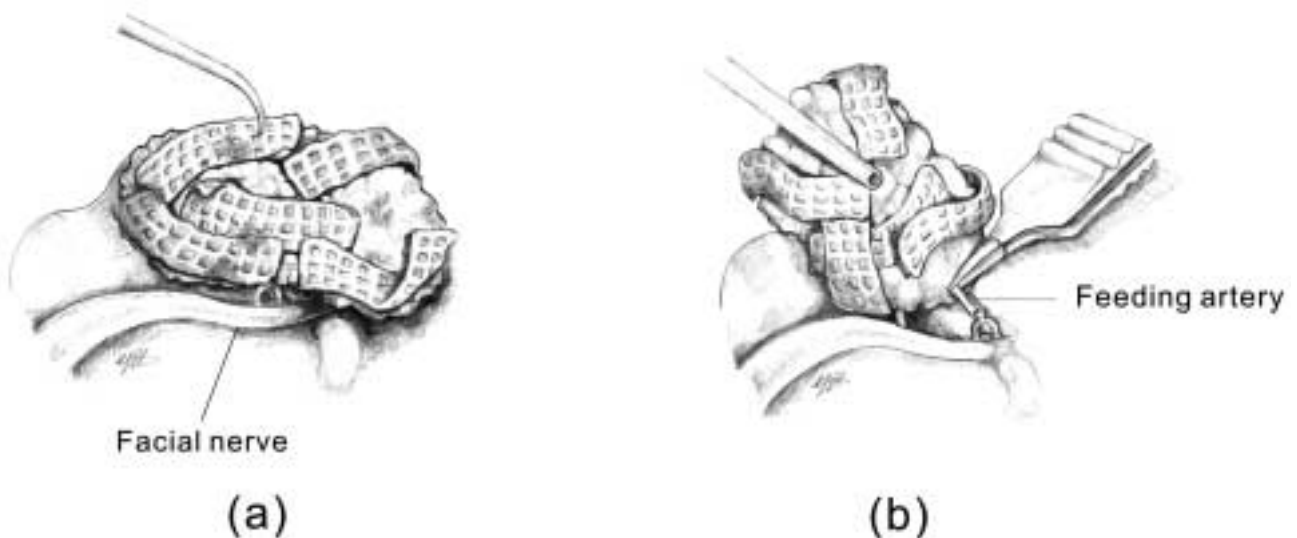


Figure 4. Wrapping and compression of the tumor by piecemeal adrenaline-soaked cotton pledgets. (b) en bloc excision of the tumor with bipolar diathermy.

are unnecessary for small GT, especially for small type A tumors^[1-3]. During the operation, whether the extensively hypervascular tumor excision should be proceeded or terminated to arrange angiography and embolization prior to next surgery was controversial to decide at that time. The frozen section played a vital role in making therapeutic decisions in our case. Nevertheless, it did not provide enough diagnostic information because of its limitations, possibly the insufficient specimen acquisition. Thus, some useful techniques regarding how to remove the tumor completely without massive bleeding were proposed intraoperatively. First, a controlled hypotensive technique (defined as a reduction of the systolic blood pressure to 80-90 mm Hg, a reduction of MAP to 50-65 mm Hg or a 30% reduction of baseline MAP) was conducted to reduce bleeding and provide a satisfactory bloodless surgical field.^[4] Furthermore, the extensive bleeding was also minimized by application of the use of canal wall-down approach, which provided a sufficient surgical field, so that the tumor volume would diminish gradually due to the vasoconstrictive effect by means of piecemeal adrenaline-soaked cotton pledgets wrapping and compression. Thus, the feeding vessels of the tumor from tympanic arteries could be recognized clearly which facilitated en bloc resection of the tumor with the aid of the bipolar diathermy. In addition, for a Fisch type B GT tumor with extensive middle ear involvement, Nadol et al. suggested that it may require a canal wall-down approach for tumor removal and an adequate meatoplasty will allow surveillance for tumor recurrence postoperatively^[5]. Therefore, for extirpation of an extensive middle ear tumor of uncertain nature with bleeding tendency, the controlled hypotensive technique and canal wall-down approach with adrenaline-soaked cotton pledgets wrapping and compression could be considered.

Conclusion

The clinical features of GT may be variable and clinicians should bear in mind the differential diagnosis of GT while encountering a middle ear mass with external auditory canal and mastoid extension. Besides, our surgical experience demonstrated that a canal wall-down approach combined with adrenaline-soaked cotton pledgets wrapping and compression under controlled hypotension without preoperative embolization is feasible for the Fisch type B GT.

Acknowledgement

We would like to give special thanks to our colleagues, Dr. Yao-Te Tsai and Dr. Po-Hung Chang. Otherwise the illustrations of Figure 3 & 4 could not have been presented in this article without their drawing endowments and computer skills.

References

1. Nadol JB Jr, McKenna MJ. Surgery of the Ear and Temporal Bone, 2nd ed. Philadelphia: Lippincott Williams & Wilkins; 2005. p. 501-521.
2. Ridder GJ, Schipper J. Paragangliomas of the head and neck: diagnosis and treatment. *Fam Cancer*. 2005; 4:55-59.
3. Tasar M, Yetiser S. Glomus tumors: therapeutic role of selective embolization. *J Craniofac Surg*. 2004; 15:497-505.
4. Degoute CS. Controlled hypotension: a guide to drug choice. *Drugs* 2007; 67:1053-1076.
5. Nadol JB Jr, Schuknecht HF. Obliteration of the mastoid in the treatment of tumors of the temporal bone. *Ann Otol Rhinol Laryngol* 1984; 93:6-12.