

Kulak Burun Bogaz Ihtis Derg 2016;26(4):238-240

Case Report / Olgu Sunumu

238

# Isolated middle ear meningioma

İzole orta kulak meningiomu

Ayşegül Batıoğlu Karaaltın, MD.,<sup>1</sup> Fatih Nesrettin Turgut, MD.,<sup>1</sup> Mehmet Yılmaz, MD.,<sup>1</sup> Şebnem Batur, MD.,<sup>2</sup> Özcan Öztürk, MD.<sup>1</sup>

<sup>1</sup>Department of Otolaryngology, İstanbul University Cerrahpaşa Medical Faculty, İstanbul, Turkey <sup>2</sup>Department of Pathology, İstanbul University Cerrahpaşa Medical Faculty, İstanbul, Turkey

#### ABSTRACT

Meningiomas are slowly progressive, benign tumors that originate from meningothelial cells. Extracranial meningiomas, especially isolated middle ear meningiomas, are very rare. In this article, we report a rare secretory type primary middle ear meningioma which was histopathologically confirmed in a 46-year-old female patient who presented with otological and neurootological symptoms.

Keywords: Chronic otitis; isolated; meningioma; middle ear.

# ÖΖ

Meningiomlar, meningotelial hücrelerden köken alan yavaş seyirli, benign tümörlerdir. Ekstrakraniyal meningiomlar, özellikle izole orta kulak meningiomları, oldukça nadirdir. Bu yazıda, otolojik ve nörootolojik semptomlarla başvuran 46 yaşındaki bir kadın hastada histopatolojik olarak teyit edilen nadir, sekretuar tipte primer orta kulak meningiomu sunuldu.

Anahtar Sözcükler: Kronik otit; izole; meningiom; orta kulak.

Meningiomas are slowly progressive, benign tumors that originate from meningothelial cells. They make up nearly 18% of primary intracranial tumors.<sup>[1]</sup> Rare extracranial meningiomas (2%) can be seen mostly in the head and neck area, especially the sinonasal area, the ear and the temporal bone.<sup>[2,3]</sup> Primary middle ear lesions without an intracranial component are very rare. In this case study, a rare secretory type primary middle ear meningioma that was histopathologically confirmed is shown in a patient with otological and neurootological symptoms.

# **CASE REPORT**

A 46-year-old female patient consulted the Otorhinolaryngology outpatient clinic with aural fullness, hearing loss and dizziness. The patient's medical history revealed that she had a ventilation tube application because of serous otitis media one year ago. Tympanogram analysis that was done at the time was reported to be type B. Otomicroscopic examination performed in our clinic showed that the right tympanic membrane was intact with a fullness that reflected a pink color. This imagery resembled a mass behind the tympanic



Available online at www.kbbihtisas.org doi: 10.5606/kbbihtisas.2016.85126 QR (Quick Response) Code Received / *Geliş tarihi:* July 12, 2015 Accepted / *Kabul tarihi:* December 28, 2015 *Correspondence / İletişim adresi:* Fatih Nesrettin Turgut, MD. İstanbul Üniversitesi Cerrahpaşa Tıp Fakültesi Kulak Burun Boğaz Hastalıkları Anabilim Dalı, 34098 Cerrahpaşa, Fatih, İstanbul, Turkey.

Tel: +90 541 - 638 06 38 e-mail (e-posta): drnftkbb@gmail.com

membrane. Nasopharnyngeal examination was normal. Other otorhinolaryngological examinations showed no pathology. Computed tomography of temporal bone revealed the right ossicular chain was slightly eroded and there was some soft tissue density in the tympanic cavity. Audiometric examination revealed that compared to pure sound average, the patient had normal levels on the left ear and a light mixed hearing loss in the right ear. Tympanoplasty under general anesthesia was performed because of these symptoms, and during the surgery some soft-tissue related to the tympanic membrane around the ossicle was observed (Figure 1). The incus was slightly eroded. Soft tissue behind the tympanic membrane and middle ear was totally excised. Type 2 tympanoplasty was completed after ossiculoplasty. A written informed consent was obtained from the patient.

Histopathology revealed a tumoral infiltration that was made up of meningothelial cells that were locally vortexed. Tumor cells stained positive for periodic acid schiff (PAS) (Figure 2) and showed intracellular eosinophilic, homogenous, round-shaped inclusions. Immunohistochemical staining showed tumor cells were positive for epithelial membrane antigen (EMA) (Figure 3) and progesterone. The case was reported to be a secretory meningioma. There were no symptoms of relapse during the postoperative eight-month follow-ups.

#### DISCUSSION

Meningiomas are slowly progressing, benign tumors that originate from dura. They make up approximately 13-26% of all primary intracranial tumors.<sup>[4]</sup> Many temporal meningioma patients with otological and neurootological symptoms have vertigo, hearing loss, tinnitus, aural fullness and ear discharge.<sup>[5,6]</sup> Our patient consulted the clinic with vertigo and aural fullness. In a case series where meningioma diagnosis was delayed due to a primary diagnosis of otitis media,<sup>[7]</sup> serious discharge attacks were found to be the general symptom after the application of a ventilation tube. Our patient had a ventilation tube applied one year ago, but she did not have the discharge symptom.

Audiological examination was reported to be in the normal range in some cases,<sup>[8]</sup> but

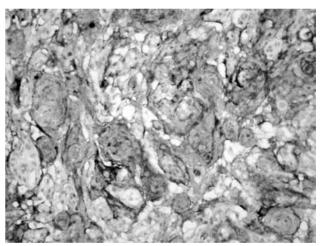
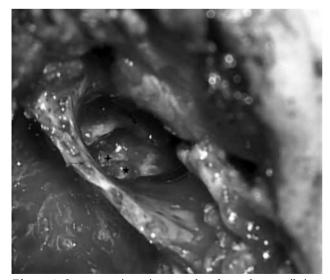


Figure 2. Secretory meningioma (H-E x 400).



*Figure 1.* Intraoperative view, marks show the manibrium mallei, stapes, posterior wall of external ear, and soft tissue (meningioma).

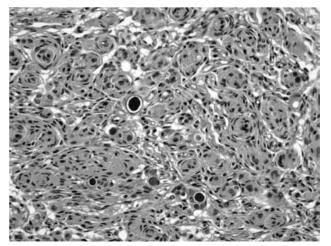


Figure 3. Secretory meningioma (PAS x 400).

was reported to be conductive hearing loss in others.<sup>[6]</sup> Our patient's audiometric examination revealed a slight mixed hearing loss. The primary examination to safely understand the size and borders of a temporal meningioma is gadolinium magnetic resonance imaging.<sup>[4]</sup> Computed tomography is a not a good method to observe meningioma but was applied to our patient who we thought to have middle ear pathology.

Meningiomas are usually benign. However, some cases may have malignant transformation. The World Health Organization classification suggests grade 1 to be benign, grade 2 to be atypical and grade 3 to be anaplastic. It was observed that transitional grade 1 meningiomas are related to extracranial meningiomas.<sup>[3]</sup> Our patient was diagnosed with a rare secretory (grade 1) meningioma after the specimen was examined histopathologically. Definitive diagnosis is usually confirmed pathologically and the treatment is total excision.

Secretory meningioma is a very rare variant and in a study the rate of incidence was reported to be 3% in all meningiomas.<sup>[9]</sup>

The most outstanding feature of secretory meningiomas is epithelial differentiation with secretory materials seen as hyaline inclusions. The intracellular, eosinophilic inclusions in meningiomas are defined as pseudopsammoma bodies. These bodies could be in different sizes, and are round-shaped deposits that are positive for PAS and that react positively for carcinoembryonic antigen, EMA and cytokeratin on immunohistochemical examination.<sup>[10]</sup> In our case, the morphological and immunohistochemical findings were similar.

Although meningioma prognoses are good, their recurrence rate ranges between 7-84%.<sup>[11]</sup> Long-term close follow-ups are important.<sup>[12]</sup> Our patient had a lesion that was mostly located around the ossicular chain and closely related to the tympanic membrane. The soft tissue was totally excised during surgery and diagnosed definitively with a pathological examination. Eight month follow-up revealed no relapse.

In conclusion, isolated middle ear meningiomas are rarely observed situations.

They can be differentiated from other middle ear pathologies by histopathological examination. Although rare, in patients with otological and neurootological symptoms that have middle ear mass lesions, meningiomas should be considered. Furthermore, diagnosed patients have to be followed-up for a long period to rule out relapse.

# **Declaration of conflicting interests**

The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

## Funding

The authors received no financial support for the research and/or authorship of this article.

## REFERENCES

- 1. Friedman CD, Costantino PD, Teitelbaum B, Berktold RE, Sisson GA Sr. Primary extracranial meningiomas of the head and neck. Laryngoscope 1990;100:41-8.
- 2. Parisier SC, Som PM, Shugar JM, Marovitz WF. The evaluation of middle ear meningiomas using computerized axial tomography. Laryngoscope 1978;88:1170-7.
- 3. D'Angelo AJ Jr, Marlowe A, Marlowe FI, Mobini J. Primary meningiomas of the middle ear: do they exist? Ear Nose Throat J 1991;70:794-8.
- 4. Whittle IR, Smith C, Navoo P, Collie D. Meningiomas Lancet 2004;363:1535-43.
- 5. Manjaly JG, Watson GM, Jones M. Primary meningioma of the middle ear. JRSM Short Rep 2011;2:92.
- 6. George M, Ikonomidis C, Pusztaszeri M, Monnier P. Primary meningioma of the middle ear: case report. J Laryngol Otol 2010;124:572-4.
- Ayache D, Trabalzini F, Bordure P, Gratacap B, Darrouzet V, Schmerber S, et al. Serous otitis media revealing temporal en plaque meningioma. Otol Neurotol 2006;27:992-8.
- 8. Jun HJ, Im GJ, Lee SH, Kwon SY, Chae SW, Jung HH, et al. An isolated middle ear meningioma. Otol Neurotol 2012;33:63-4.
- Alguacil-Garcia A, Pettigrew NM, Sima AA. Secretory meningioma. A distinct subtype of meningioma. Am J Surg Pathol 1986;10:102-11.
- 10. Sav A, Soylemezoglu F, Ozer F, Pamir N, Kullu S, Ekicioglu G. Secretory meningioma a conventional histochemical study of six cases. Turkish Neurosurgery 1991:2:10-3.
- 11. Thompson LD, Bouffard JP, Sandberg GD, Mena H. Primary ear and temporal bone meningiomas: a clinicopathologic study of 36 cases with a review of the literature. Mod Pathol 2003;16:236-45.
- 12. Kumar G, Basu S, Sen P, Kamal SA, Jiskoot PM. Ectopic meningioma: a case report with a literature review. Eur Arch Otorhinolaryngol 2006;263:426-9.