



Two cases with unusual mycetoma localizations in upper respiratory system

Üst solunum yolunda alışılmadık miçetoma yerleşimli iki olgu

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Fungus balls or mycetomas are extramucosal and noninvasive accumulations of degenerating fungal hyphae. In head and neck they may localized most frequently in the paranasal sinuses, especially in the maxillary sinus. These indolent infections are mostly caused by *Aspergillus spp.* In this article, we present two rare fungus ball cases: one occurring in an automastoidectomy cavity of a temporal bone and the other in a concha bullosa. Typically, both patients admitted with vague symptoms consistent with chronic infection resistant to conventional antibacterial medication.

Key Words: Concha bullosa; fungus ball; mycetoma; temporal bone.

Fungus topları veya miçetomalar, dejenerasyon gösteren fungal hiflerin ekstramukozal ve noninvazif birikimlerinden oluşur. Baş boyun bölgesinde en sık paranasal sinüslerde, özellikle maksiller sinüste yerleşim gösterirler. Bu sinsi enfeksiyonlara çoğunlukla *Aspergillus suşları* neden olmaktadır. Bu makalede biri temporal kemiğin otomastoidektomi boşluğunda ve diğeri konka bulloza içerisinde yerleşimli iki nadir fungus topu olgusu sunuldu. Tipik olarak her iki hasta da konvansiyonel antibakteriyel tedaviye yanıt vermeyen ve kronik enfeksiyonla uyumlu muğlak semptomlar sergilemekteydi.

Anahtar Sözcükler: Konka bulloza; fungus topu; miçetoma; temporal kemik.

Fungus balls or mycetomas are extramucosal accumulations of degenerating fungal hyphae especially within chronically inflamed paranasal sinuses.^[1] We report two rare cases of mycetomas, one occurring in an automastoidectomy cavity and the other in a concha bullosa.

CASE REPORT

Case 1- A 50-year-old male admitted with progressive hearing loss in the previous 10 years, bilateral otorrhea especially prominent in the right ear since childhood and vertigo accompanied

by falling attacks that had started six months before. On physical examination he had a retracted tympanic membrane and a posterosuperior cholesteatoma in the left ear and a draining right ear with an adhesive tympanic membrane and granulation tissue. Pure tone audiometry revealed a 60 dB conductive hearing loss (CHL) in the left and 55 dB CHL in the right ear. Temporal bone computed tomography (CT) demonstrated bilateral automastoidectomy cavities and posterior external auditory canal wall destruction, inner cortical mastoid bone destruction, thinning of the right

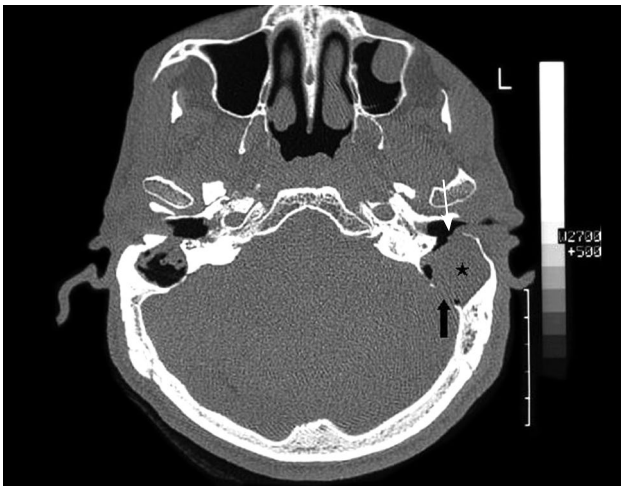


Figure 1. Axial temporal bone computed tomography shows temporal bone fungus ball (asterisk), erosion of inner mastoid cortex (arrow) and destruction of the medial portion of posterior ear canal wall (white arrow).

otic capsule and a round shaped mass in the right mastoid cavity with soft tissue density which was considered to be granulation tissue or a giant cholesteatoma (Figure 1, 2). A surgery was planned for the right ear. Following a retroauricular incision, a cholesteatoma matrix was detected just below the overly thinned outer mastoid cortex. After incision of the cholesteatoma matrix, instead of debris, a soft, brown, mucopurulent, cheesy tissue was found. Cholesteatoma matrix was overlying the mastoid cavity. There was a bone defect exposing the posterior fossa dura. The mastoidectomy cavity was cleaned of cholesteatoma and the cheesy tissue. A tragal cartilage graft was placed between malleus and stapes. The open attic was also grafted with cartilage and a closed tympanum was

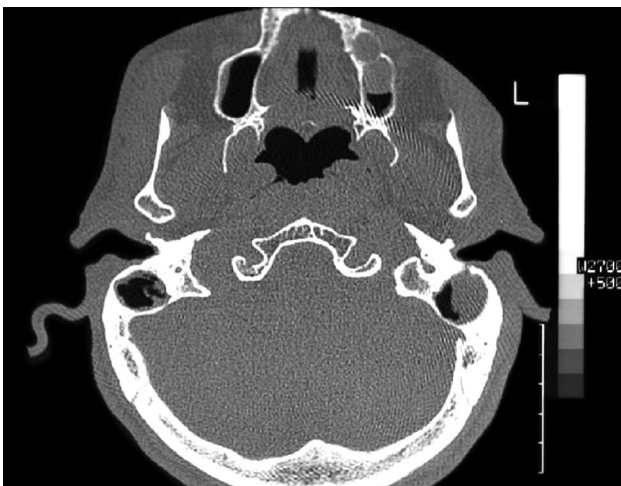


Figure 2. An inferior level scan shows bilateral auto-mastoidectomy cavities.

achieved by an underlay temporalis fascia graft. Pathology reported the presence of hyphae that led to the diagnosis of fungus ball.

Case 2– A 15-year-old female was seen with complaints of nasal obstruction, snoring and frequent bouts of headache for two years. She did not have apneas or recurring sinusitis. On ear, nose and throat (ENT) examination a left-sided septal deviation and a polypoid mass filling the right nasal cavity were noted. Paranasal CT revealed a giant right middle turbinate with hyperdense areas in the middle turbinate suggesting a fungal infection of a concha bullosa (Figure 3). T1 weighted magnetic resonance imaging demonstrated isointense middle concha cavity (Figure 4). During surgery when the middle turbinate was excised, a great amount of mucopurulent secretions and clay-like substrate were aspirated. The entire excess wall of the middle turbinate was excised. Pathology reported fungal elements taken from the specimen with no invasion of the conchal bone. The patient had no immunocompromised condition or steroid use history. In the six-week follow-up the nasal mucosa healed with no recurrence of fungal infection.

DISCUSSION

Fungus balls are extramucosal accumulations of degenerating hyphae.^[1] In the head and neck they are almost always seen in the paranasal sinuses and especially in the maxillary sinus. Almost

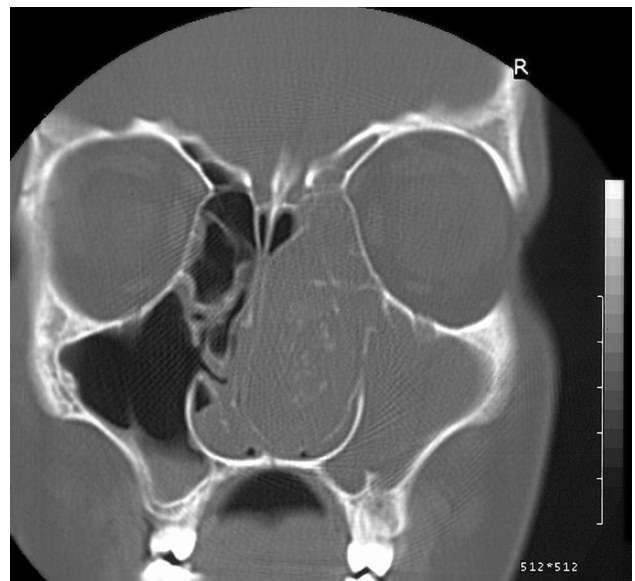


Figure 3. Coronal paranasal sinus computed tomography demonstrating a giant middle concha at the right side with calcified foci.



Figure 4. T₁ weighted magnetic resonance imaging demonstrating heterogeneous izointense bullous middle concha cavity.

all paranasal sinus fungus balls are caused by *Aspergillus* spp.

Surgical removal of a fungus ball in a paranasal sinus is curative and usually there is no need for antifungals unless there is an accompanying invasive form.^[1] Radiologically they may cause bony thickening or sclerosis of the walls, calcifications and bone destruction.^[2] Pathologically they are extramucosal and noninvasive.

Fungal infections of the head and neck may occur in the paranasal sinuses, oral cavity, pharynx, larynx, esophagus and ear. Mycoses of the ear predominantly appear in the form of external auditory canal involvement i.e. otomycosis. Fungal malignant otitis externa and fungal mastoiditis are rarely reported.^[3,4]

Furthermore fungal DNA was also demonstrated in the middle ear fluid of patients with otitis media effusion (OME) suggesting a possible role in its pathogenesis.

In case 1, due to the co-occurrence of cholesteatoma and fungus ball, the reason for automastoidectomy cannot be stated for certain, because both pathologies are capable of causing bone destruction. We propose that the most possible explanation in this case is expansion of fungus-colonized cholesteatoma sac. The CT findings of the case reflect a retrospective diagnosis of a temporal bone mycetoma. The spherical appearance of the lesion in the preoperative CT scans in addition to the numerous intact aerated air cells surrounding the lesion implies a fairly rapid de novo expansion of the lesion rather than slowly progressive debris accumulation.

Although pathology reported hyphae in both cases, which gomori methenamine silver and periodic acid schiff highlighted stains, fungal growth in cultures was not observed. This failure of demonstration of fungal growth is probably due to the lack of viability of fungi that was clearly demonstrated in the series of Ferreiro et al., wherein out of 22 paranasal sinus fungus ball cultures, 17 showed no growth.^[2]

In conclusion, fungus balls or mycetomas may occur in automastoidectomy cavities of the temporal bone and in concha bullosae.

REFERENCES

1. deShazo RD, O'Brien M, Chapin K, Soto-Aguilar M, Swain R, Lyons M, et al. Criteria for the diagnosis of sinus mycetoma. *J Allergy Clin Immunol* 1997;99:475-85.
2. Ferreiro JA, Carlson BA, Cody DT 3rd. Paranasal sinus fungus balls. *Head Neck* 1997;19:481-6.
3. Yao M, Messner AH. Fungal malignant otitis externa due to *Scedosporium apiospermum*. *Ann Otol Rhinol Laryngol* 2001;110:377-80.
4. Ohki M, Ito K, Ishimoto S. Fungal mastoiditis in an immunocompetent adult. *Eur Arch Otorhinolaryngol* 2001;258:106-8.