

Giant cell granuloma of the temporomandibular joint: a report of the two cases

Temporomandibüler eklemin dev hücreli granüloması: İki olgu sunumu

Ahmet Mutlu, MD., Fatih Sarı, MD., Murat Öztürk, MD., Mete İşeri, MD.

Department of Otolaryngology, Medical Faculty of Kocaeli University, Kocaeli, Turkey

Giant cell granulomas (GCG) are non-neoplastic lesions that are mostly found in the oral cavity, maxilla and mandible and are mostly common in young ages (20-40 years).^[1,2] The temporal region is an extraordinary place for these benign lesions. To date, four cases of the temporomandibular joint (TMJ) GCG have been reported in the English literature, and the main pathophysiology has not been elucidated.^[3] According to their clinical aspects, the required clinical treatment is surgical excision with clean margins.^[4] In this letter, we report the clinical evaluation of two TMJ GCG cases that destroyed the temporal bone.

Case 1- A 38-year-old female was referred with the complaint of one-year progressive hearing loss, tinnitus, and TMJ malocclusion. There was no history of chronic otitis, trauma, hyperparathyroidism or medical intervention. On examination, a red colored non-pulsatile solid mass obliterated the external auditory canal on the right ear (Figure 1). The pure tone audiometer displayed severe mixed hearing loss detected on the right ear (R: 76 dB L: 36 dB). No facial paralysis was detected. Temporal bone computed tomography (CT) showed destruction of the petrous apex, and mastoiditis on the right ear. Contrast cranial magnetic

resonance (MR) T₁-T₂ weighted images showed a 30x20x20 mm hyperintense lesion with destruction of the mastoid bone, petrous apex, and TMJ (Figure 2). The patient underwent infratemporal type B approach and TMJ

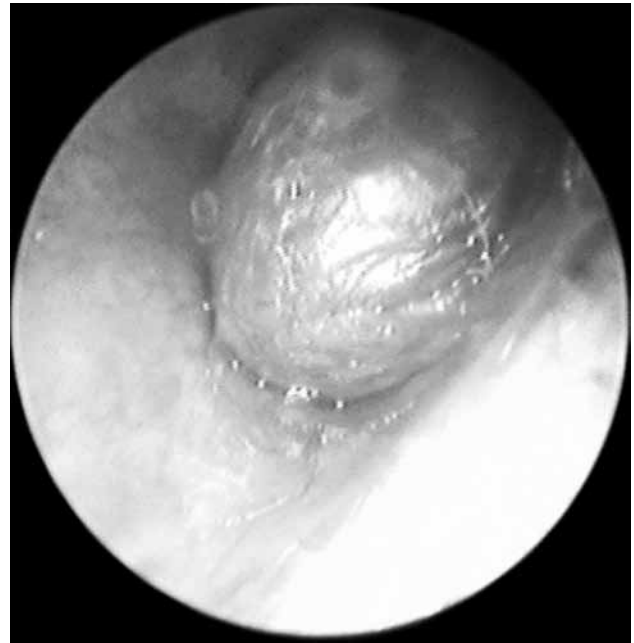


Figure 1. A red colored non-pulsatile solid mass obliterates the external auditory canal of the right ear.



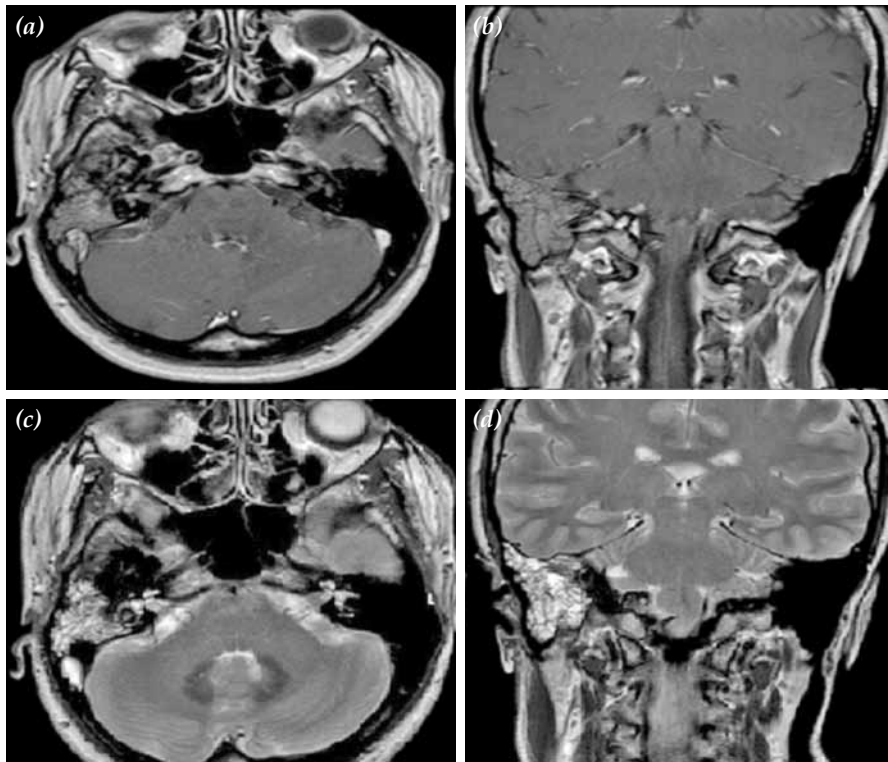


Figure 2. Magnetic resonance images. (a) Axial section, T₁-weighted images, (b) coronal section, T₁-weighted images, (c) axial section, T₂-weighted images (d) coronal section, T₂-weighted images showing a 30x20x20 mm hyperintense lesion with destruction of the mastoid bone, petrous apex, and temporomandibular joint.

resection because of the TMJ lesion. No dural invasion was detected. The defect was filled with abdominal fat graft. Postoperatively no facial paralysis or TMJ malocclusion were encountered. At 24-months follow-up, there is no symptom of recurrence or residual lesion.

Case 2- A 46-year-old male consulted with a complaint of chronic otitis externa, progressive hearing loss, and preauricular mass on the left ear. On examination a blue colored solid lesion expanding from the anterior wall obliterating the external auditory canal and purulent otorrhea were noted. The temporal CT revealed a TMJ originated lesion destroying the external auditory canal and middle ear (Figure 3). The temporal MR T₁-T₂ weighted images showed a 25x25x10 mm hyperintense lesion. The patient underwent infratemporal type B approach as well and the mass was resected with the mandibular condyle with clear surgical margins. The facial nerve and dura were kept intact, and on postoperative follow-up there was no symptom of facial paralysis or cerebrospinal

fluid (CSF) leakage. On 24-months follow-up, no recurrence or residual lesions were detected.

Multi-nucleated giant cells are characterized on histopathological studies and hemosiderin deposits may be determined (Figure 4). Due to the clinical features of the lesion, aggressive giant cell tumor of the long bone and Brown tumor must be considered in the differential diagnosis. On pathological surveys giant cell tumor of the long bone have widespread mitotic activity and more giant cells than GCG.^[4] Brown tumor is hardly distinguished from GCG, however high blood levels of parathyroid hormones can help to detect Brown tumors.^[5] The TMJ has to be considered as a potential location for GCG, and the glenoid fossa surface and condyle should be evaluated on radiological examinations. The preferred treatment is surgical resection. On surgery, the TMJ periosteum has to be removed, and the glenoid fossa surface and dural invasion must be checked to avoid recurrences. Dural or periosteal invasion may be clinical findings associated with a malignant process. In case

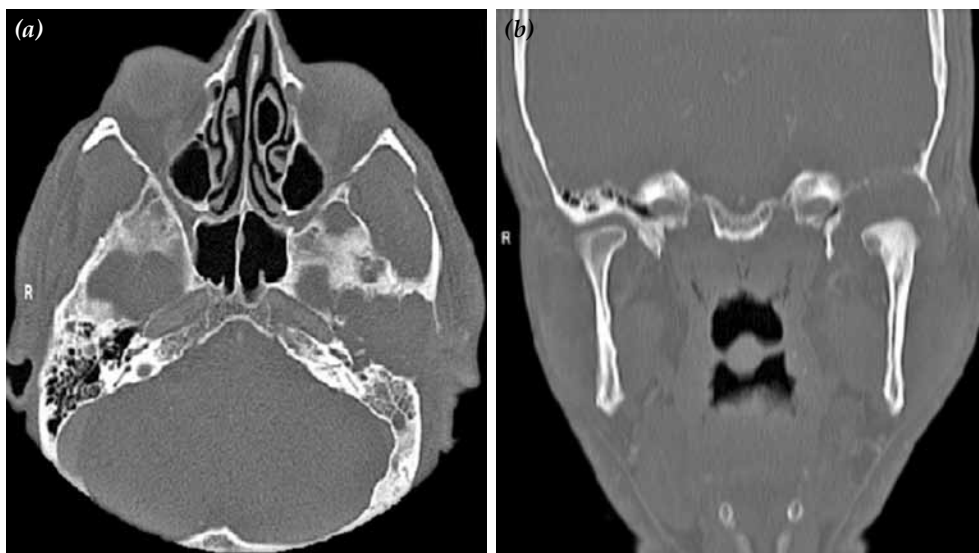


Figure 3. Temporal computed tomography. (a) Axial section and (b) coronal section revealed a temporomandibular joint originated lesion with destruction of the external auditory canal and middle ear.

of incomplete resection or comorbid disorders that prevent the primary surgery, systemic calcitonin therapy, intralesional alendronate and corticosteroid injections may be considered to obtain satisfactory results and control this dilemma. However, effectiveness and duration of these therapies remain unclear.^[2,6,7] We preferred surgery on both cases because of inadequacy of the pathological diagnosis, and the inadequately-known effect of medical therapy on joint-originated lesions. Radiotherapy is another treatment method to manage these benign lesions yet the application is controversial due to the

potential risk of sarcomatous induction.^[4,6] Kaban et al.^[8] reported an effective antiangiogenic therapy for aggressive giant cell granulomas and showed that interferon-2 alpha may help to maintain disease control. In our cases our histopathological surveys did not raise suspicion of aggressive GCG and we did not practice other additional therapies.

Declaration of conflicting interests

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REFERENCES

1. Van Buren N, Buchmann L, Gurgel R, Shelton C. Giant cell granuloma of the temporal bone. *Otol Neurotol* 2014;35:211-2.
2. Ma Y, Liu L, Huang D, Han D, Wang J, Wu W, et al. Diagnosis and treatment of giant cell granuloma of the temporal bone: Report of eight cases. *Acta Otolaryngol* 2012;132:657-62.
3. Jadu FM, Pharoah MJ, Lee L, Baker GI, Allidina A. Central giant cell granuloma of the mandibular condyle: a case report and review of the literature. *Dentomaxillofac Radiol* 2011;40:60-4.
4. Menge M, Maier W, Feuerhake F, Kaminsky J, Pfeiffer J. Giant cell reparative granuloma of the temporal bone. *Acta Neurochir (Wien)* 2009;151:397-9.

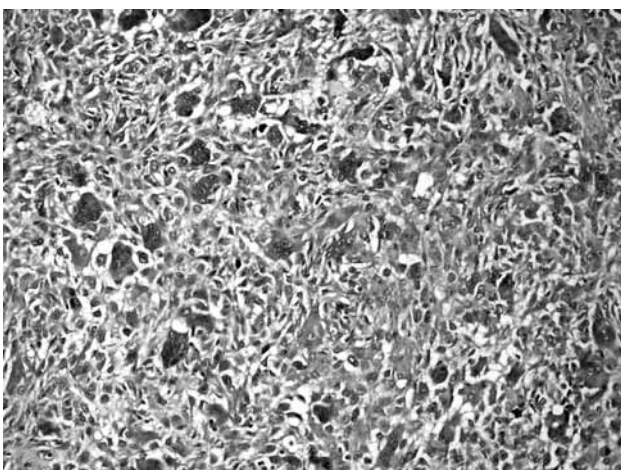


Figure 4. Histopathological examination image. Multi-nucleated giant cells have been widely placed (H-E x 200).

5. Praveen AH, Thriveni R. Maxillary and mandibular hyperparathyroidism. Maxillary and mandibular hyperparathyroidism. *Natl J Maxillofac Surg* 2012;3:51-4.
6. Souter MA, Bird PA, Worthington JP. Giant cell reparative granuloma of the temporal bone treated with calcitonin. *Otol Neurotol* 2006;27:999-1002.
7. da Silva NG, Carreira AS, Pedreira EN, Tuji FM, Ortega KL, de Jesus Viana Pinheiro J. Treatment of central giant cell lesions using bisphosphonates with intralesional corticosteroid injections. *Head Face Med* 2012;8:23.
8. Kaban LB, Troulis MJ, Ebb D, August M, Hornicek FJ, Dodson TB. Antiangiogenic therapy with interferon alpha for giant cell lesions of the jaws. *J Oral Maxillofac Surg* 2002;60:1103-11.