

# PERIPHERAL OSTEOID LOOK LIKE CALCIFICATION OF THE BODY OF ZYGOMATIC BONE: CASE REPORT OF AN UNUSUAL LOCALIZATION

## ПЕРИФЕРНА ОСТЕОИДНА-НАЛИК ФОРМАЦИЈА НА ТЕЛО НА ЗИГОМАТИЧНА КОСКА: ПРИКАЗ НА СЛУЧАЈ СО НЕСПЕЦИФИЧНА ЛОКАЛИЗАЦИЈА

Naumovski S.<sup>1</sup>, Benedetti A.<sup>2</sup>, Stamatovski A.<sup>1</sup>

<sup>1</sup>Trainee in maxillofacial surgery, DDM, Clinic for maxillofacial surgery Skopje, Faculty of Dental Medicine, <sup>2</sup>DDM, MD, DDS, PhD, MFS, Clinic for maxillofacial surgery Skopje, Faculty of Dental Medicine

### Abstract

Osteomas of zygomatic bone are especially rare benign osseous tumors. Although this tumour has a considerable incidence, there are few reports with large samples depending on this anatomical region. Three theories have been proposed and various possible etiologies have been discussed in the literature, but symptoms are not always apparent and specific. **The purpose** of this article is to present the clinical, radiographic, surgical finding and management of unspecific right zygomatic osteoma in a 35-year old woman following diagnostic dilemmas. The lesion was treated surgically without complications and the patient made a complete recovery.

**Keywords:** zygomatic bone, osteoma, jaw, osteoid osteoma

### Апстракт

Остеомите на зигоматична коска претставуваат ретки и бенигни туморски формации. За инциденцата на овие коскени тумори и тоа на зигоматична регија, последните литературни податоци говорат за објавени само неколку статии и трудови за оваа патологија. Постојат три теории и повеќе етиолошки фактори за тоа како настануваат остеомите. Симптоматологијата не секогаш ја презентира во целост почетната спорорастечка фаза на ваков бениген тумор со потекло од коскено ткиво. **Целта** на овој труд е да се прикаже клиничката слика, дијагностички методи и третман на избор кај зигоматичен остеома заедно со диференцијална дијагноза кај 35-годишна пациентка. **Клучни зборови:** зигоматична коска, остеома, вилица, остеоиден остеома

### Introduction

Osteoma is a slow growing benign osseous lesion that comes from well-differentiated mature bone tissue with three variants of central, peripheral and extra skeletal. They are often asymptomatic and usually continuously growing, and occur most often in adolescents and predominantly in females with a mean age at diagnosis of about 46 years, and rarely can be found in maxillary bones. However, osteoma of the zygoma is rare<sup>1-5</sup>.

The etiology of osteomas remains unknown. The first author who recognized this pathological entity was Jaffe in 1935<sup>1</sup>. Osteoma in craniofacial skeleton is detected on routine x-ray examinations or when they reach a large size, they can produce swelling and asymmetry, pain, dysfunction, jaw deviation etc<sup>5-7</sup>. Radiologically, osteo-

mas are presented as a well-defined radiopaque mass with density similar to the normal bone. Genetic testing and colonoscopy are indicated in adolescents and young adults, because of the Gardner's syndrome (GS) and the development of colorectal adenocarcinoma<sup>2-5</sup>.

Differential diagnosis should include osteomyelitis (e.g. Brodie abscess), osteoblastoma: >2 cm in size, fibrous dysplasia, cortical desmoid, fibroma, osteitis, odontoma, enostosis (bone island), reactive sclerosis around with osteolytic lesion.

Surgical removal of osteomas depends on the location, extent, and existence and is indicated when the lesion is symptomatic, actively growing, or causing esthetic disfigurement, trismus, malocclusion and functional impairment.

We present a rare case of a peripheral osteoma of the right zygomatic bone in an elderly woman.

## Case presentation

A 35-year-old female was referred to our clinic presented with a painless firm mass in the right malar area in April 2019, firstly noticing this lesion 3 years earlier and she became concerned as it slowly enlarged. There was no trauma or infection, and her medical history did not contain any known pathology of the intestines.

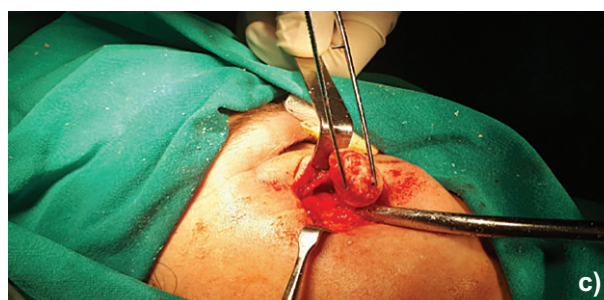
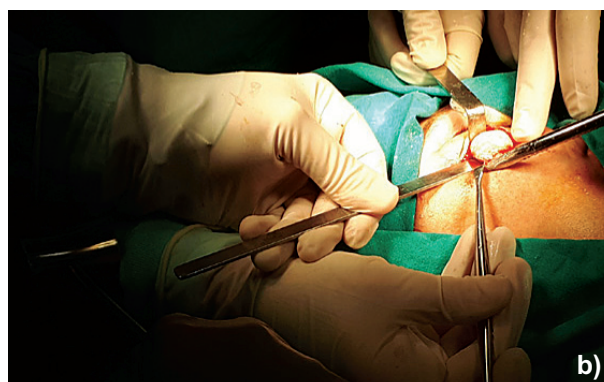
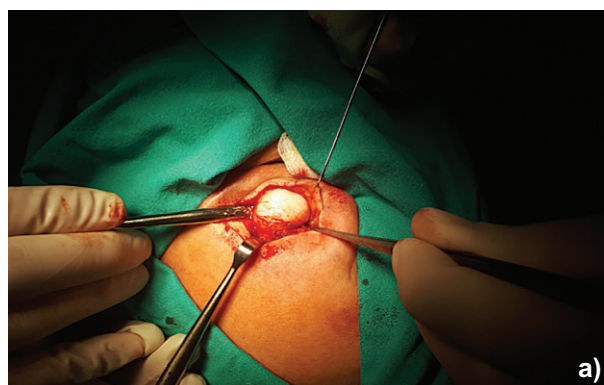
Extra orally, we revealed solitary, smooth, firm, bony asymptomatic mushroom-like mass over the right zygoma, with deformity of the face measuring approximately 3.5 cm in diameter (Figure 1). The overlying skin was normal in color with no signs of inflammation. There were no ophthalmologic symptoms, headache or facial pain in our patient. The routine blood investigations were also within the normal limits. The regional lymph nodes were non-palpable. The lesion was bony-hard on palpation.

A computed tomography (CT) scan revealed unilateral well - defined lobulated mass protruding from the outer cortex of the right zygoma to the adjacent anterior cheek



**Figure 1.** Clinical preoperative view showing swelling on the right side of the face

about 2.01cm×1.57cm×2.32cm in antero-posterior, transverse and cranio-caudal dimensions respectively. Due to increasing cosmetic and asymmetry reasons of the face, a decision was made to remove the tumor under general anesthesia with preservation of nearby structures. After incision, a complete view of the lesion was obtained (Figure 2a, 2b). The tissue was reflected by blunt dissection above the right zygomatic bone with skin flap and nerve stimulator to detect the branch of facial nerve during surgical procedure (Figure 2c, 2d). The tumor was completely removed (Figure 3). No elevation of temperature at the affected site and no pus discharge were observed. The patient was discharged home 3 days after surgery. Histopathologic examination revealed a well-circumscribed mass composed of dense lamellar bone with osteoclastic activity. Histological examination assessed an



**Figure 2.** Intraoperative view showing various steps in removal of the osteoma followed by a clinical photo after resection and recontouring the cortical bone

osteoid osteoma with normal bone architecture. The post-operative 6 month follow up period was uneventful without complications or recurrence and the patient made a



**Figure 3.** Macroscopic pathologic specimen excised showing the lobulated bony mass

complete recovery. Prognosis is excellent except for the rare cases of malignant transformation.

## Discussion

Osteomas of cranio-maxillo-facial region are extremely uncommon, especially in the malar bone anatomical side with unclear etiology<sup>3-6</sup>. They are composed of mature compact or cancellous bone and sometimes may be found in association with other diseases such as polyps, fibromatous lesions of the connective tissue and the Gardner's syndrome (familial adenomatous polyposis) 7-10. Its incidence is rare in the jaws and the mandible is more affected than the maxilla. A literature review identified only few previously documented cases of zygomatic bone osteoma. Osteoma can arise at any age, but is more frequently seen between the fourth and fifth decade of life.

N. Larrea-Oyarbide et al.,<sup>3</sup> in their retrospective study of 106 patients reported that 132 osteomas of the craniomaxillofacial region were found to be diagnosed between 1986 and 2003, with mean age of 50 years.

Several authors proposed that osteomas are often asymptomatic and usually slow growing and may cause different complications depending on the part of the central nervous or visual systems<sup>2, 5-7</sup>.

On the other side Johann AC et al.,<sup>5</sup> showed 69 well-documented cases of peripheral osteoma with peripheral type and the mandible was the most frequently affected side. The radiopaque focal mass with rarefaction and reactive bone formation is typical for this type of tumors, but even diagnostic tools (CT scan or MRI) von Chamier G et al.,<sup>7</sup> suggested that the initial changes are often uncharacteristic and can cause further delay in proper diagnosis because of the dense lamellae arranged in layers, and clinically should be differentiated from several pathologies.

Radiographic x-ray investigations such as occlusal radiograph, panoramic radiograph or Computed Tomography (CT) with 3D reconstruction are proposal.

We believe that the intraoral approach has lower risks of facial injury and scarring, and because of the poor visibility, we decided to use direct extraoral facial incision required osteotomy for adequate surgical exposure, a good view of the lesion, complete tumor resection with satisfactory esthetic results and normal bone architecture. The patient is under follow – up and no signs of recurrence have been observed so far.

## Conclusion

Osteomas appear in patients older than 35 years, and usually do not present clinical symptoms of pain, headache, neuralgia or paresthesia. The present case was not part of the Gardner syndrome, but association of osteoma with the Gardner's syndrome must always be kept in mind. Proper history, physical and radiological examination is necessary to identify the location and extent of the osteoma. Complete surgical excision is the treatment of choice. Positioning of a titanium plate was not necessary, and the tumor was completely removed from the anterior wall of the zygoma. The postoperative result provided good morphological, functional and aesthetic outcome with no evidence of recurrence.

## Conflict of interest

The authors declare that they have no conflicts of interest.

### Source of Support:

Nil.

### Ethical disclosures

**Protection of human and animal subjects:** The authors declare that no experiments were performed on humans or animals for this investigation.

**Confidentiality of data:** The authors declare that they have followed the protocols of their work centre on the publication of patient data and that the patient included in the study have received sufficient information and have given their informed consent in writing to participate in that study.

## Reference

1. H. L. Jaffe, "Osteoid osteoma: a benign osteoblastic tumor composed of osteoid and atypical region," *Archives of Surgery*, vol. 31, no. 5, pp. 709–728, 1935.
2. Dae Young Kim, Kap Sung Oh. A Rare Case of Peripheral Osteoma of the Zygoma. *Arch Plast Surg*. 2015 Jan; 42(1): 103–105. Published online 2015 Jan 14. doi: 10.5999/aps.2015.42.1.103.
3. Larrea-Oyarbide N, Valmaseda-Castellón E, Berini-Aytés L, Gay-Escoda C. Osteomas of the craniofacial region. Review of 106 cases. *J Oral Pathol Med*. 2008 Jan;37(1):38-42.

- 
4. Wanyura H, Kamiński A, Stopa Z. Treatment of osteomas located between the anterior cranial base and the face. *J Craniomaxillofac Surg.* 2005 Aug;33(4):267-75.
  5. Johann AC, de Freitas JB, de Aguiar MC, de Araújo NS, Mesquita RA. Peripheral osteoma of the mandible: case report and review of the literature. *J Craniomaxillofac Surg.* 2005 Aug;33(4):276-81.
  6. V. I. Akinmoladun, A. O. Akadiri, and A. A. Olusanya, "Peripheral osteoma of the zygoma: literature review and case presentation," *African Journal of Medicine and Medical Sciences*, vol. 36, no. 4, pp. 381–383, 2007.
  7. von Chamier G, Holl-Wieden A, Stenzel M, Raab P, Darge K, Girschick HJ, Beer M. Pitfalls in diagnostics of hip pain: osteoid osteoma and osteoblastoma. *Rheumatol Int.* 2010 Jan;30(3):395-400. doi: 10.1007/s00296-009-0943-2. Epub 2009 May 15.
  8. Rodriguez Y Baena R1, Rizzo S, Fiandrino G, Lupi S, Galioto S. Mandibular traumatic peripheral osteoma: a case report. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2011 Dec;112(6):e44-8. doi: 10.1016/j.tripleo.2011.05.006. Epub 2011 Sep 8.
  9. F. K. Horikawa, R. R. de Freitas, F. A. Maciel, and A. J. Gonçalves, "Peripheral osteoma of the maxillofacial region: a study of 10 cases," *Brazilian Journal of Otorhinolaryngology*, vol. 78, no. 5, pp. 38–43, 2012.
  10. T. Starch-Jensen, "Peripheral solitary osteoma of the zygomatic arch: a case report and literature review," *The Open Dentistry Journal*, vol. 11, no. 1, pp. 120–125, 2017