

# **RESEARCH ARTICLE**

# INTRAOSSEOUS CAVERNOUS HEMANGIOMA OF THE ZYGOMA: A CASE REPORT

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# Manuscript Info

#### Abstract

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#### Key words:-

Cavernous Hemangioma, Zygomatic Bone, Vascular Malformation, Intraosseous Hemangioma The incidence of intraosseous hemangiomas is low; they represent less than 1% of all bone tumors. The most common locations are the calvaria and vertebrae. Lesions affecting facial bones are rare and can involve the maxillae, mandibulae, nasals, and zygomatics. In zygomatic location it's a slow-growing tumor that primarily affects adult women. Herein, we report a case of intraosseous hemangioma of the zygomatic bone in a man aged of 55 years old. The diagnosis of intraosseous hemangioma was made following radiological examination due to the characteristic imaging findings and confirmed after surgical excision by anatomopathological exam.

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#### **Introduction:-**

Bone hemangiomas are rare vascular lesions, accounting for 0.5 to 1% of all primary bone tumors. Zygomatic localization is exceptional and mostly involved bones are vertebra and the calvaria in maxillofacial region.Imagery plays a crucial role in positive diagnosis, but his presentation is nonspecific and can mime other bones tumors. Therefore, anatomopathological study appears to be indispensable for a certitude diagnosis. Surgical resection is the main treatment. Herein, we report a case of intraosseous hemangioma in the zygomatic bone.

## **Case Report:-**

Mr. B.M, a 55-year-old male, with a history of vitiligo and mental disorders under treatment; consults our structure for a slight painless right jugal swelling evolving for two years with a history of local trauma.Palpationexam reveals a hard, immobile mass attached to the zygomatic bone, measuring approximately 2,5 cm in its largest dimension, with a normal appearance of soft tissues and skin in regards.

A facial CT scan confirms an intraosseous expansive process in the rounded malar bone, well-demarcated with trabecular sunburst appearance, causing small interruptions in the bone cortices at certain points. Topographically, it extends from the left zygomatic arch downwards into the maxillary sinus, slightly bulging into its lumen. Upwards and inwards, it protrudes into the infero-external quadrant of the orbit, respecting the orbital contents. Outwards, it bulges into the soft jugal tissues, altering the skin contour without affecting the subcutaneous fat. (Figure 1)

The overall appearance strongly supports a diagnosis of osseous hemangioma.

**Corresponding Author:- Habimana Alain François** Address:- Department of Maxillofacial Surgery, University Hospital Center Ibn Sina of Rabat Morocco. A surgical resection under general anesthesia, was decided and performed through an endobuccal approach, by milling with a ball burr and subsequent regularization of the bone surface. No reconstruction was performed since there was no aesthetic impact after the excision. (Figure 2)

After what, the specimen was sent for a histopathological examination that confirmed spongy bone tissue harboring cavernous vascular clefts, supporting the diagnosis of intraosseous cavernous hemangioma in the zygomatic bone.

Postoperative recovery was uneventful, resulting in a satisfactory aesthetic outcome and no recurrence during a 2-year follow-up.

#### **Discussion:-**

Intraosseous hemangioma is a slow-growing, rare tumor originating from newly formed blood vessels.

The first case reported of zygomatic bone hemangioma dates to 1950 by Schoenfield (9).

In 2017, according to Powers DB and al., only 49 cases were reported in the literature and most of them as case reports. (13)

It predominantly affects vertebrae and the cranial vault, with maxillofacial locations being even rarer (13).

The peak incidence occurs is around the fourth decade, predominantly in females (6)(7). The etiology remains uncertain;traumatic or congenital causes have been reported to be associated with the development of such lesions (11).

Clinically, they manifest as a firm, painless mass with slow progressive growth, causing facial asymmetry. Pain, if present can be the most common symptom, followed by ocular manifestations related to mass effect, such as dystopia, exorbitism, ptosis, and interference with extraocular muscle movements (13).

Histologically, the tumor can appear under four variant types: capillary type, cavernous type, mixed variant, and scirrhous type (13). In facial bone involvement hemangiomas are mostly cavernous (12).

CT- scan is the most useful imaging technique, by providing detailed trabecular and cortical information for accurate lesion topography and determining its benign, non-invasive nature, along with the absence of extension into adjacent soft tissues. The radiographic appearance is often described as a *'honeycomb'*, *'soap bubble'*, *or 'sunburst'*(13).

Although initial concerns may suggest osteosarcoma, careful examination reveals intact cortical borders, emphasizing the benign nature of these lesions (15). Contrast injection shows significant tumor enhancement (2).

Management of primary intraosseous hemangiomas of the zygoma depends on both symptoms and aesthetic considerations and varies based on the location and extent of the lesion (13).

In contrast to venous malformations of soft tissues for which various therapeutic options have been described, complete surgical resection remains the treatment of choice for primary intraosseous hemangiomas (15)(16)(17)(18), thereby minimizing the risk of recurrence.

Surgical access to the infra-temporal fossa is challenging due to the anatomical complexity and difficulty in accessing this region (3)(4)(5).

En-bloc resection with adequate bone margins appears to minimize the risk of massive bleeding (19)(20)(21). Consequently, despite the vascular nature of these lesions, hemorrhage is a rare complication (13).

Reconstruction can be achieved using autogenous bone or calvarial grafts or by resorting to alloplastic implant prostheses, such as PEEK implants (13).

## **Conclusion:-**

Bone hemangiomas are benign vascular tumors rarely occurring in the maxillofacial region, with zygomatic localization presenting a significant aesthetic challenge for maxillofacial surgeons. CT facilitates precise lesion mapping, confirming its benign, non-invasive nature and the absence of extension into adjacent soft tissues. Given the zygoma's importance in facial aesthetics, reconstruction should be considered, if necessary, in cases of osseous hemangiomas.

## Iconography



Figure 1:- Facial CT scan axial, coronal and 3D Reconstruction, Lesional Process in the body of right zygomatic region.



Figure 2:- Facial and <sup>3</sup>/<sub>4</sub> view of the patient after a year after surgery.

#### **References:-**

- 1. Skiker\*, F. Benchakroun, M. Fikri, M.R. El Hassani, M. Jiddane.Localisationsrares de l'hémangiomeintraosseux; Feuillets de radiologie2012; 52:322-325
- 2. Raji A, Essaadi M, Laraqui NZ, Janah A, Kadiri F, Touhami M, et al. L'angiomemandibulaire:a propos d'un cas et revue de la litte rature. Sem Hop Paris 1998; 74:1249–54.
- 3. Kornfehl J, Gstöttner W, Kontrus M, Sedivy R. Transpalatine ex- cision of a cavernous hemangioma of the infra-temporal fossa. Eur Arch Otorhinolaryngol1996; 253:172-5.
- 4. Prades JM, Timoshenko A, Merzougui N, Martin C. A ca- daveric study of a combined trans-mandibular and trans- zygomatic approach to the infra-temporal fossa. Surg Radiol Anat 2003; 25:180-7.
- 5. Sarac S, Koybasi S, Kaya S. Transmaxillary excision of a rare cavernous hemangioma of the infra-temporal fossa. Ear Nose Throat J 2000; 79:448-9,452.
- 6. Ozdemir R, Alagoz S, Cagri Uysal A, Erkin Unlu R, OrtakT, Sensoz O. Intraosseous hemangioma of the mandible: a case report and review of the literature. J Craniofac Surg 2002;13: 38–43.
- Werle AH, Kirse DJ, Nopper AJ, Toboada EM. Osseous heman- gioma of the maxilla in an infant. Otolaryngol Head Neck Surg 2003; 128:906–9.
- 8. Chagnon S , Valle' e C , Ble' ry M , Chevrot A. Hémangiome et angiomatose diffuse. Encycl Med Chir Radiodiagnostic-Neuror- adiologie-Appareil locomoteur 1992;IV:1-19 [31489 A10].
- 9. Schofield AL. Primary hemangioma of the malar bone. Br J Plast Surg 1950;3(2):136-140
- Kaya B, Işılgan SE, Cerkez C, Otrakçı V, Serel S. Intraosseous cavernous hemangioma: a rare presentation in maxilla. Eplasty 2014;14: e35
- 11. Park BH, Hwang E, Kim CH. Primary intraosseous hemangioma in the frontal bone. Arch Plast Surg 2013;40(3):283-285
- 12. Aykan A, Keleş MK, Bayram Y, et al. Hemangioma of the zygomatic bone. J Craniofac Surg2016;27(2): e200e202
- Powers DB, Fisher E, Erdmann D. Zygomatic Intraosseous Hemangioma: Case Report and Literature Review. Craniomaxillofac Trauma Reconstr. 2017 Mar;10(1):1-10. doi:10.1055/s-0036-1592087. Epub 2016 Nov 4. PMID: 28210401; PMCID: PMC5309129.
- 14. Moore S L, Chun J K, Mitre S A, Som P M. Intraosseous hemangioma of the zygoma: CT and MR findings. AJNR Am J Neuroradiol. 2001;22(7):1383–1385.
- 15. Defazio M V, Kassira W, Camison L. et al. Intraosseous venous malformations of the zygoma:clarification of misconceptions regarding diagnosis and management. Ann Plast Surg. 2014;72(3):323–327.
- 16. Cheng N C, Lai D M, Hsie M H, Liao S L, Chen Y B. Intraosseous hemangiomas of the facial bone. PlastReconstr Surg. 2006;117(7):2366–2372.
- 17. Yu M S, Kim H C, Jang Y J. Removal of a nasal bone intraosseous venous malformation and primary reconstruction of the surgical defect using open rhinoplasty. Int J Oral Maxillofac Surg. 2010;39(4):394–396.
- 18. Walker E A Jr, McHenry L C. Primary hemangioma of the zygoma. Arch Otolaryngol. 1965; 81:199–203.
- 19. Koybasi S, Saydam L, Kutluay L. Intraosseous hemangioma of the zygoma. Am J Otolaryngol. 2003;24(3):194–197.
- 20. Gómez E, González T, Arias J, Lasaletta L. Three-dimensional reconstruction after removal of zygomatic intraosseous hemangioma. Oral Maxillofac Surg. 2008;12(3):159–162.
- 21. de Ponte F S, Bottini D J, Valentini V. [Surgical treatment of hemangioma of bones of the orbito-zygomatic region] Minerva Stomatol. 1994;43(7–8):365–372.