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Intraosseous venous malformation of the zygoma: A case report and literature review

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ABSTRACT

INTRODUCTION: Venous malformations of the zygoma are rare. Historically, venous malformations have been misrepresented as "hemangiomas". The International Society for the Study of Vascular Anomaly (ISSVA) classification is a reasonable classification that leads to appropriate clinical diagnosis and treatment strategies. Collaboration between surgeons, radiologists, and pathologists is necessary for accurate diagnosis and management.

PRESENTATION OF CASE: We present here a case of an IOVM in a 59-year-old woman who was treated with a multidisciplinary approach. Superselective arteriography and embolization were effective for diagnosis as well as for prevention of large hemorrhage during surgery. En-bloc resection of the zygoma was performed within hours after embolization and autologous calvarial bone graft was used for primary reconstruction.

DISCUSSION: We performed a literature review consisting of reviewing 52 cases of IOVM of the zygoma discussing optimal material for reconstruction of the defect for intraosseous venous malformation of the zygoma nationally and internationally.

CONCLUSION: The combination of surgery and preoperative angiography makes it possible to prevent high risk of hemorrhage. For primary reconstruction of the zygoma, use of autologous calvarial bone can maintain the volume and reconstruct the natural malar contour.

1. Introduction

Venous malformations of the bone of the maxillofacial region are rare. They account for less than 1% of all bony "tumors" [1,2,3] and are most frequently described in the vertebral column and calvarium [2]. The maxilla and mandible are the most commonly affected bones of the facial skeleton [3], followed by the zygoma, the orbit, and the condyle [2]. Fifty two cases of venous malformation (VM) of the zygoma have been reported in the literature [12,3]. Intraosseous venous malformation (IOVM) presents a rare yet unique clinical challenge to the surgeon. The keys for treatment are: (1) hemorrhage control, (2) en-bloc resection of the bony lesion including the normal bone, and (3) reconstruction after resection. Because of significant hemorrhage risk, intraosseous vascular anomalies can be life-threatening entities. Twenty five deaths by spontaneous hemorrhage have been reported in the literature [1]. In these literature, even the small lesions of VM may have significant hemorrhage risk. We present here a case of IOVM of the zygoma. Preoperative angiography was performed to confirm the diagnosis followed by embolization to reduce the risk of bleeding during the operation. A good cosmetic result was achieved using autologous calvarial bone for reconstruction of the zygoma defect.

2. Presentation of a case

A 59-year-old female presented with a 3-month history of progressive painless swelling of the left cheek. A hard, immobile mass of the malar eminence was palpable (Fig. 1A). Computed tomography (CT) showed a round, well-defined expansile bony lesion measuring 5 cm in the left zygoma. The honeycomb lesion was also evident on 3-dimensional CT (3DCT) (Fig. 1B). Posteriorly, the lesion tapered into the anterior zygomatic arch. There were neither periosteal lesions nor any associated soft tissue lesions. The mass had an overall intermediate T1 signal intensity and a high T2 signal intensity on magnetic resonance imaging (MRI). Within the mass were areas of no signal that corresponded to the trabeculae seen on the CT study. No extra-osseous soft tissue component was apparent. On biopsy, significant hemorrhage was caused. The pathology revealed the lesion to be a cavernous hemangioma.

Preoperative superselective angiography and embolization were performed. Selective bilateral external carotid artery angiograms displayed markedly hypertrophied branches of the left facial artery and left internal maxillary artery (Fig. 2A). After microcatheter embolization, en-bloc resection of the zygoma including

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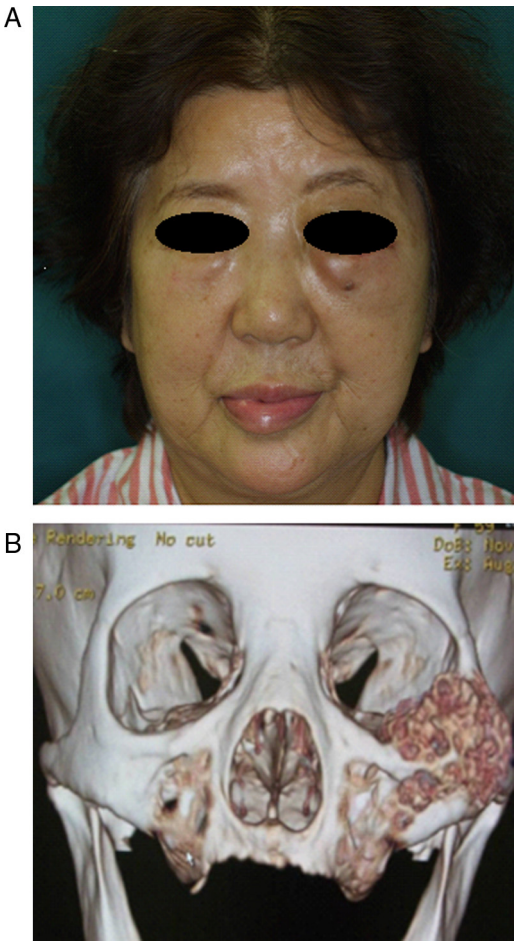

Fig. 1. (A) Preoperative view. (B) 3DCT scan image shows a honeycomb lesion in the left zygoma.
The lesion is a mass of mixed density over the malar eminence of the zygomatic bone involving the lateral orbital wall on the left side.

the VM was performed with a subcilial and intraoral approach to avoid postoperative facial nerve palsy caused by preauricular approach. The total blood loss was approximately 500 ml. Anatomical reconstruction of the zygoma and orbital floor was performed by using a 5 × 5 cm split parietal calvarial bone for the zygoma and a split iliac bone for the orbital floor. The split calvarial bone was taken primarily using a surgical chisel. The resulting calvarial bone donor site was repaired using hydroxyapatite bone cement.

Histology of the affected area (hematoxylin and eosin staining) revealed thin-walled, enlarged vascular channels with a single layer of flat, quiescent endothelium between bony trabeculae. The endothelium showed no signs of proliferation, mitotic figures, atypia, or tufting, and the bony margins did not show any sign of vascular anomaly.

Follow-up at 6 months demonstrated no significant deformity of the left zygoma, with good contour of the midface on clinical examination (Fig. 2B). CT scan at 6 month postoperatively, revealed volume maintenance and anatomical continuity of the calvarial bone grafts (Fig. 3). After over 3 years, the patient appears no recurrence or deformity of the left zygoma.

3. Discussion

A literature review of IOVM of the zygoma is shown in Table 1. Hemangiomas (Hs) and cavernous hemangiomas (CHs) are included types of IOVM. The incidence of intraosseous venous malformations of the zygoma occurs in a female to male ratio of 4.5:1. In 7% of cases, preoperative arterial embolization followed by total

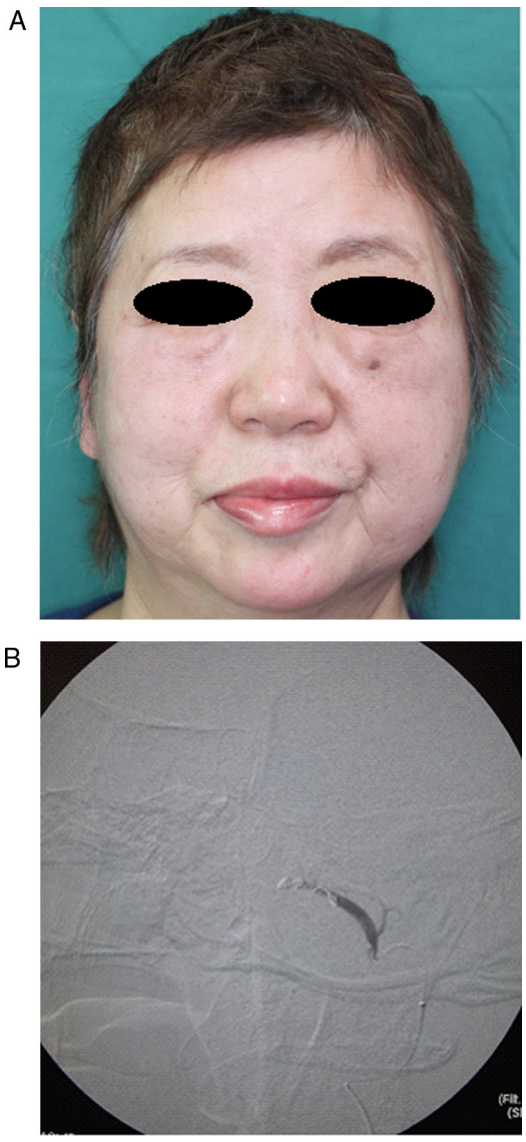

Fig. 2. (A) Lateral view of the selective left internal maxillary arteriogram before embolization. Homogenous staining was seen in the circle. The left internal maxillary artery was feeding the lesion. (B) Patient at 6 months postoperative.

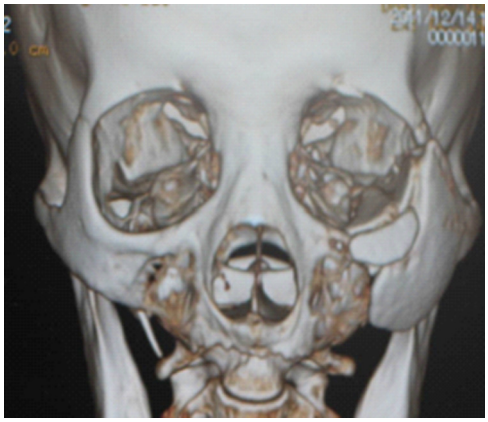

Fig. 3. 3DCT scan imaging 6 months postoperatively.