Oral Oncology Reports 8 (2023) 100114

Contents lists available at ScienceDirect

Oral Oncology Reports

journal homepage: www.journals.elsevier.com/oral-oncology-reports

Fronto-orbital intraosseous hemangioma with skull base erosion: When a relative "simple" disease could require a "complex" approach

ARTICLE INFO

Keywords
Osseous hemangioma
CAD/CAM reconstruction
Vascular tumors
Head and Neck cancer

ABSTRACT

Osseous hemangiomas are rare benign vascular bone tumors usually involving skull bones. Sometimes it is difficult to exclude a malignant tumor, since they are vascularized lesions associated with bone erosion and to perform an incisional biopsy in some cases can be challenging for the risk of bleeding. We present a case of osseous hemangioma involving fronto-orbital region associated with dura mater exposure by extensive anterior cranial fossa erosion, that underwent radical surgical excision after pre-operative sclerotization. CAD/CAM technology reconstruction permitted to guarantee an adequate aesthetic-functional result and to reduce operative time.

Dear Editor;

Vascular anomalies are congenital abnormalities of vascular development that often involve the head and neck region [1]. Primarily separated into vascular tumors and vascular malformations by the International Society for the Study of Vascular Anomalies (ISSVA) classification, they include a wide range of phenotypically and histologically different lesions [2,3].

Osseous hemangiomas usually present as rare benign vascular bone lesions (1 %), and in most cases skull bones are involved (80 %) [4,5]. Histological diagnosis is defined by the presence of massive osteolysis and bone replacement by blood vascular spaces filled with red blood cells and fibrous hyalinized stroma containing proliferating capillaries [6]. Given its rarity and variable clinical appearance, osseous angiomatosis is not easily diagnosed. Imaging plays an important role for diagnostic informations and therapeutic planning in most arduous cases, but in many occasions surgery may be necessary according to tumor size, tissue invasion and location, availability, patient age and aesthetic appearance [7].

Only few reports can be found about management of craniofacial intraosseous hemangiomas; due to the fact that they present as vascularized lesions associated with bone erosion, sometimes it is difficult to exclude a malignant tumor. On the other side, to perform an incisional biopsy in some cases can be challenging for the risk of haemorrhage.

Recently, a 44 years-old Caucasian male presented at our centre referring a progressive left eye ptosis due to a painless mass involving the superior margin of the orbit; displacement of ocular globe in absence of diplopia was present. Previous history was unremarkable for diseases. CT scan showed the presence of a focal expansive osteolytic lesion localized in the left frontal region and at the level of the left anterosuperior orbital wall and anterior skull base leading to osseous remodelling with bony extensive erosion and thinning (Fig. 1A). Biopsy was done and ossifying angioma was suspected. MRI confirmed the orbital cavity involvement and eyeball contiguity relations (Fig. 1B). It was decided to perform surgical resection after pre-operative sclerotizing agents administration to prevent a possible bleeding and loss of blood during surgery [2,7] Computer aided design/computer assisted manufacturing (CAD/CAM) and 3-dimensionally (3D) printed models and devices were utilized as surgery and reconstruction support [8,9].

Bicoronal approach was done to expose supra-orbital margin and the resection was carried out by using 3D printed osteotomy guides (Fig. 1C). Extensive involvement of the orbital bony cortical and almost total erosion of the anterior cranial fossa (ASB) exposing the dura mater has been shown; a galea flap was opted for ASB reconstruction. In this case the use of 'bioprinting' of tissue engineered scaffolds is the gold standard in osseous defect reconstruction: patient specific bioactive bone substitute developed for bone integration and osteogenesis was utilized (Fig. 1D). Postoperative period was un-eventful; definitive histopathological examination was consistent with previous hypothesis confirmation of osseous hemangioma.

Even if osseous hemangiomas are typically asymptomatic benign vascular tumors that grow slowly and malignant transformation is very rare [10], their localisation in some occasions could have a crucial role in prognosis and therapeutic planning. In addition, in some cases initial imaging and, occasionally, rapid changes in symptoms can lead to suspect of malignant tumors. In this case, due to the involvement of fronto-orbital region associated with dura mater exposure by extensive ABS erosion, even in the presence of a benign lesion, sclerotization was not sufficient to resolve the pathology, and it was decided to perform surgical excision. Obviously, due to the localisation, it was mandatory to guarantee an adeguate aesthetic-functional result, and CAD/CAM technology permit us also to reduce operative time.

It is clear that in those cases it is fundamental to perform an adequate surgical planning with a multidisciplinary approach.

Funding source

This study did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

https://doi.org/10.1016/j.oor.2023.100114
Received 20 October 2023; Accepted 29 October 2023
Available online 11 November 2023

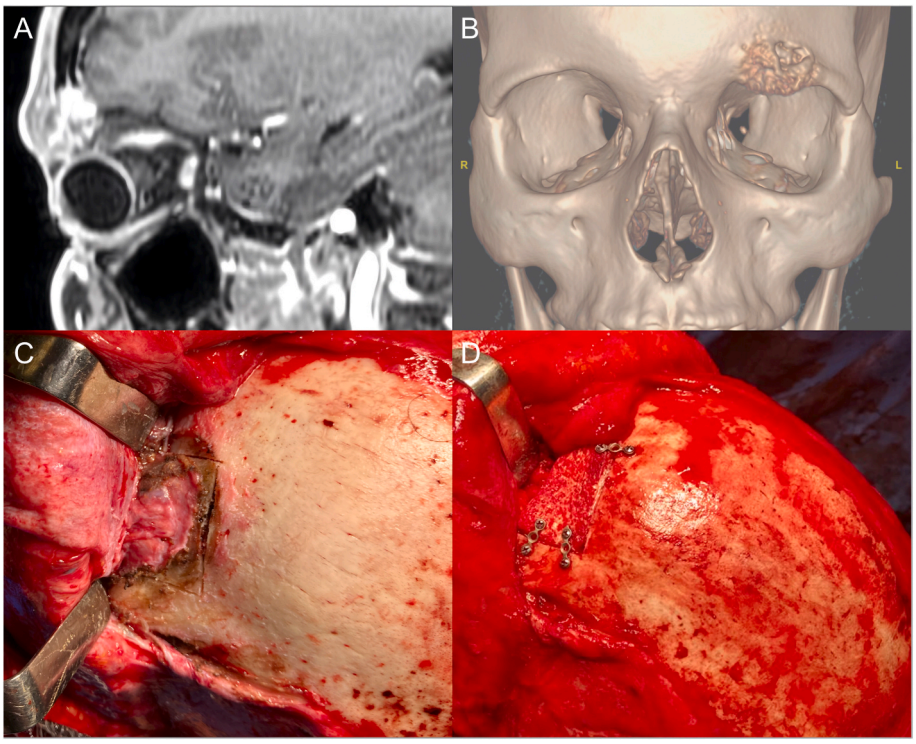

Fig. 1. A) pre-operative MRI Volumetric T1-MPRAGE at presentation, sagittal view; B) pre-operative 3D CT scan reconstruction, frontal view; C) intraoperative view after lesion sclerotization (surgical margins were demarcated by using a template); D) intraoperative view after reconstruction.

Ethical approval

The methods were carried out in accordance with the approved guidelines.

Informed consent

The patient provided written informed consent.

Contribution Author(s)

Study concepts: AB.
Study design: VT.
Data acquisition: MD.
Quality control of data and algorithms: MC.
Data analysis and interpretation: GP.
Statistical analysis: PP.
Manuscript preparation: DD, AD.
Manuscript editing: FD.
Manuscript review: VV.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Acknowledgment

None.

References

[1] Alsuwailem A, Myer 4th CM, Chaudry G. Vascular anomalies of the head and neck. Semin Pediatr Surg 2020;29:150968. https://doi.org/10.1016/j.sempedsurg.2020.150968.

[2] Cortivo FD, Diallo R, Preudhomme R, Coulibaly A, Salami A, Benateau H. Exclusive surgical approach for facial arteriovenous malformations in precarious conditions. J Stomatol Oral Maxillofac Surg 2022;123:e549–55. https://doi.org/10.1016/j.jormas.2022.01.008.

[3] Mariatos G, Gorgoulis VG, Laskaris G, Kittas C. Epithelioid hemangioma (angiolymphoid hyperplasia with eosinophilia) in the oral mucosa. A case report and review of the literature. Oral Oncol 1999;35:435–8. https://doi.org/10.1016/s1368-8375(99)00006-8.

[4] Yao K, Tang F, Min L, Zhou Y, Tu C. Multifocal intraosseous hemangioma: a case report. Medicine (Baltim) 2019;98:e14001. https://doi.org/10.1097/MD.0000000000014001.

[5] Weinandt M, Legras A, Mordant P, Le Pimpec Barthes F. Chest wall resection for multifocal osseous haemangioma. Interact Cardiovasc Thorac Surg 2016;22:233–4. https://doi.org/10.1093/icvts/ivv321.

[6] Malik R, Malik R, Tandon S, Tandon P. Skeletal angiomatosis - rare cause of bone destruction: a case report with review of literature. Indian J Pathol Microbiol 2008;51:515–8. https://doi.org/10.4103/0377-4929.43745.

[7] Romano A, Tavanti F, Rossi Espagnet MC, et al. The role of time-resolved imaging of contrast kinetics (TRICKS) magnetic resonance angiography (MRA) in the evaluation of head-neck vascular anomalies: a preliminary experience. Dento-maxillofacial Radiol 2015;44:20140302. https://doi.org/10.1259/dmfr.20140302.

[8] Nyirjesy SC, Heller M, von Windheim N, et al. The role of computer aided design/computer assisted manufacturing (CAD/CAM) and 3- dimensional printing in head and neck oncologic surgery: a review and future directions. Oral Oncol 2022;132:105976. https://doi.org/10.1016/j.oraloncology.2022.105976.

[9] Terenzi V, Della Monaca M, Raponi I, et al. MRONJ and ORNJ: when a single letter leads to substantial differences. Oral Oncol 2020;110:104817. https://doi.org/10.1016/j.oraloncology.2020.104817.

[10] Yelken Kendirci M, Ünsal G, Özcan İ, Soluk Tekkeşin M. A rare localization of intraosseous hemangioma: case report. J Adv Res Health Sci 2022;5:118–21. https://doi.org/10.26650/JARHS2022-1093263.

Andrea Battisti, Fiorenza Dal Cortivo*
Odontostomatological and Maxillofacial Sciences Department, "Sapienza" University, Policlinico Umberto I. Viale del Policlinico, 155, Rome, Italy

Mario Corona
Vascular and Interventional Radiology Unit, Radiology, Oncology and Pathology Department, "Sapienza", University of Rome, Viale Regina Elena 324, 00161, Roma, Italy

Valentina Terenzi
Surgical Sciences Department, University of Rome Tor Vergata, Rome, Italy

Marco Della Monaca, Paolo Priore, Danilo Di Giorgio, Alberto Dell'Aquila
Odontostomatological and Maxillofacial Sciences Department, "Sapienza" University, Policlinico Umberto I. Viale del Policlinico, 155, Rome, Italy

Giulio Pagliuca
ENT Unit, Santa Maria Goretti Hospital, AUSL Latina, Italy

Valentino Valentini
Odontostomatological and Maxillofacial Sciences Department, "Sapienza" University, Policlinico Umberto I. Viale del Policlinico, 155, Rome, Italy

* Corresponding author. Via Allegri 20, 37035 San Giovanni Ilarione, Verona, Italy.
E-mail address: fiorenza.dalcortivo@gmail.com (F. Dal Cortivo).