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Case Report

Hemangioma of the maxillary sinus: A benign and rare cause of epistaxis [☆]

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ARTICLE INFO

Article history:

Received 27 March 2022

Revised 23 April 2022

Accepted 28 April 2022

Keywords:

Hemangioma

Maxillary sinus

Vascular tumor

Computed tomography

Magnetic resonance imaging

ABSTRACT

Hemangioma of the facial sinuses is a rare pathology, and given the lack of clinical specificity, the differential diagnosis with a malignant lesion often arises. We report the case of a 32-year-old patient who consulted for recurrent epistaxis of moderate severity. The pre-operative diagnosis of a hemangioma of the left maxillary sinus was based on computed tomography and magnetic resonance imaging data, confirmed by the anatomopathological study of the surgical specimen, preceded by an embolization that facilitated the endoscopic surgical excision.

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Introduction

Hemangioma is a benign vascular tumor, which rarely occurs in the facial sinuses. They represent only 10% of cervico-facial localizations [1,2]. It is manifested by nasal obstruction, and recurrent epistaxis. This clinical feature, similar to other pathologies, including malignant pathology, makes the differential diagnosis difficult, based mainly on imaging. Pre-operative diagnosis is important for surgical planning.

Clinical observation

We report the case of a 32-year-old patient, who consulted for recurrent epistaxis of moderate severity. Her history of the disease goes back to childhood when the patient presented unilateral epistaxis, of low abundance, which stops spontaneously, worsened 4 months ago, becoming of great abundance, associated with facial pain.

[☆] Competing Interests: The authors declare no conflict of interest.

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<https://doi.org/10.1016/j.radcr.2022.04.062>

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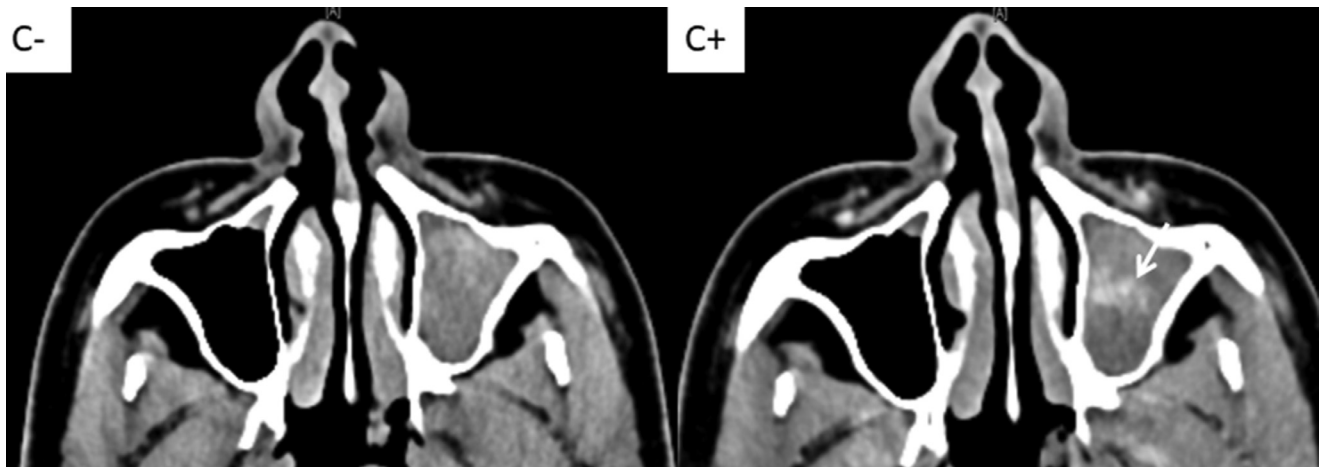


Fig. 1 – CT scan of the facial sinuses in axial section showing almost complete hematic filling of the left maxillary sinus, with an intense enhancement after injection testifying to the presence of an underlying bleeding lesion.

The computed tomography (CT) scan of the facial sinuses showed an almost complete hematic filling of the left maxillary sinus, with a heterogeneous enhancement testifying to the presence of an underlying bleeding lesion, probably benign given the absence of erosion or bone remodeling (Fig. 1). The magnetic resonance imaging (MRI) complement allowed for a better characterization of the left maxillary intrasinus lesion, measuring about 1,5 cm × 2 cm in large diameter. Presenting in hyposignal T1, heterogeneous hypersignal T2, surrounded by hyposignal T2 border, without diffusion restriction, and intensely enhanced after contrast, with an attachment zone at the level of the internal wall of the maxillary sinus. Its benign nature is highly probable, evoking a sinus hemangioma (Figs. 2 and 3).

Arteriography confirmed the hypervascular nature of the lesion, showing a vascular blush in projection of the roof of the left maxillary sinus, found after selective catheterization of the left sphenopalatine artery. This was followed by preoperative embolization by coils of the left sphenopalatine artery, with disappearance of the vascular blush at control (Fig. 4).

The postoperative follow-up after complete endonasal surgical removal of the tumor was normal. And the postoperative CT scan showed the total disappearance of the left sinus intramaxillary vascular lesion, with the embolization material centered on the left sphenopalatine artery (Figs. 5-7).

Discussion

Hemangiomas are benign vascular tumors, related to a hyperplastic proliferation of endothelial cells. Histologically, there is the capillary type hemangioma, the cavernous and the mixed form, depending on the size of the dominant vessel. Capillary hemangioma is the most common type consisting of capillary-sized vessels bordered with a flattened epithelium. They are found especially in children, characterized by their superficial localization in the skin or oral mucosa. They usu-

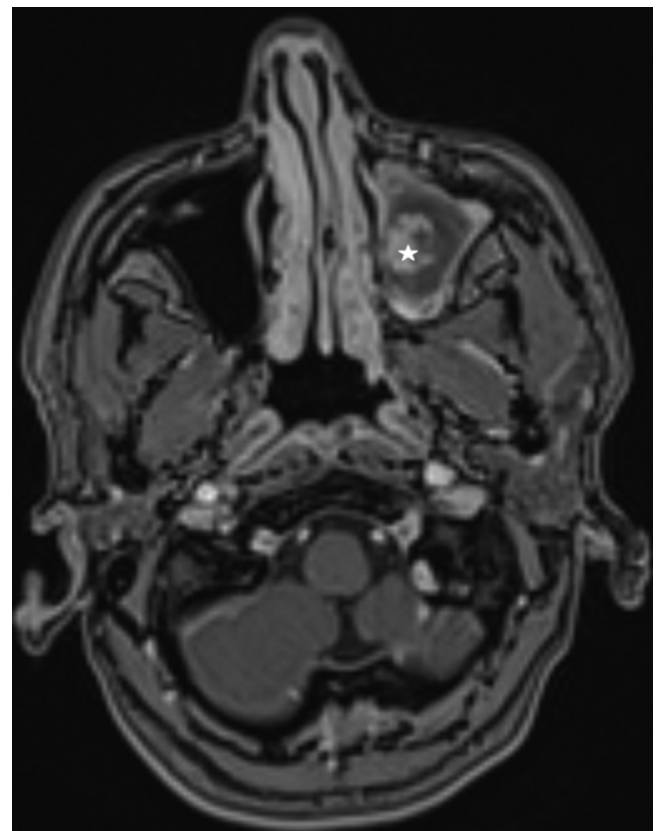


Fig. 2 – MRI in axial sequence T1 injected showing the left maxillary intrasinus lesion, intensely enhanced after contrast, well limited, suggestive a sinus hemangioma.

ally regress spontaneously by the age of 6-12 years. Cavernous hemangiomas are composed of large vascular spaces bordered by endothelium, which occur in adulthood, and do not regress spontaneously [3]. Localization in the sinuses is very

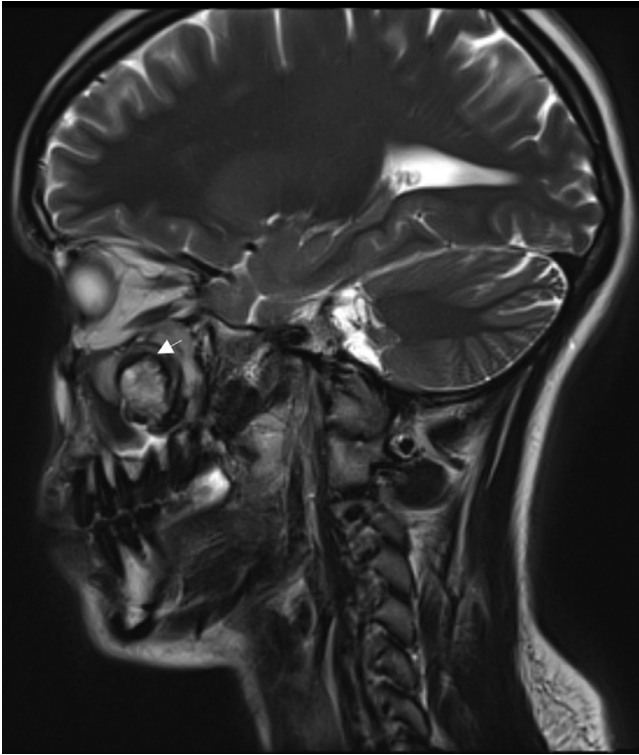


Fig. 3 – MRI in sagittal sequence T2 showing the lesion in heterogeneous hypersignal, surrounded by T2 hyposignal border.

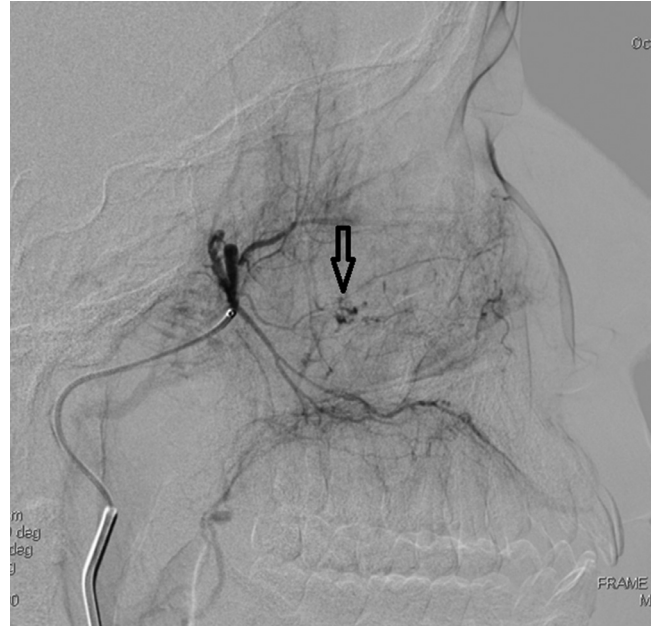


Fig. 4 – Arteriography showing a vascular blush in projection of the left maxillary sinus roof.

rare, only 10% of cervico-facial localizations [2]. These tumors evolve slowly, with a local aggressive potential by mass effect. They may be responsible for erosions and bone remodeling, posing the problem of differential diagnosis with malignant tumors.

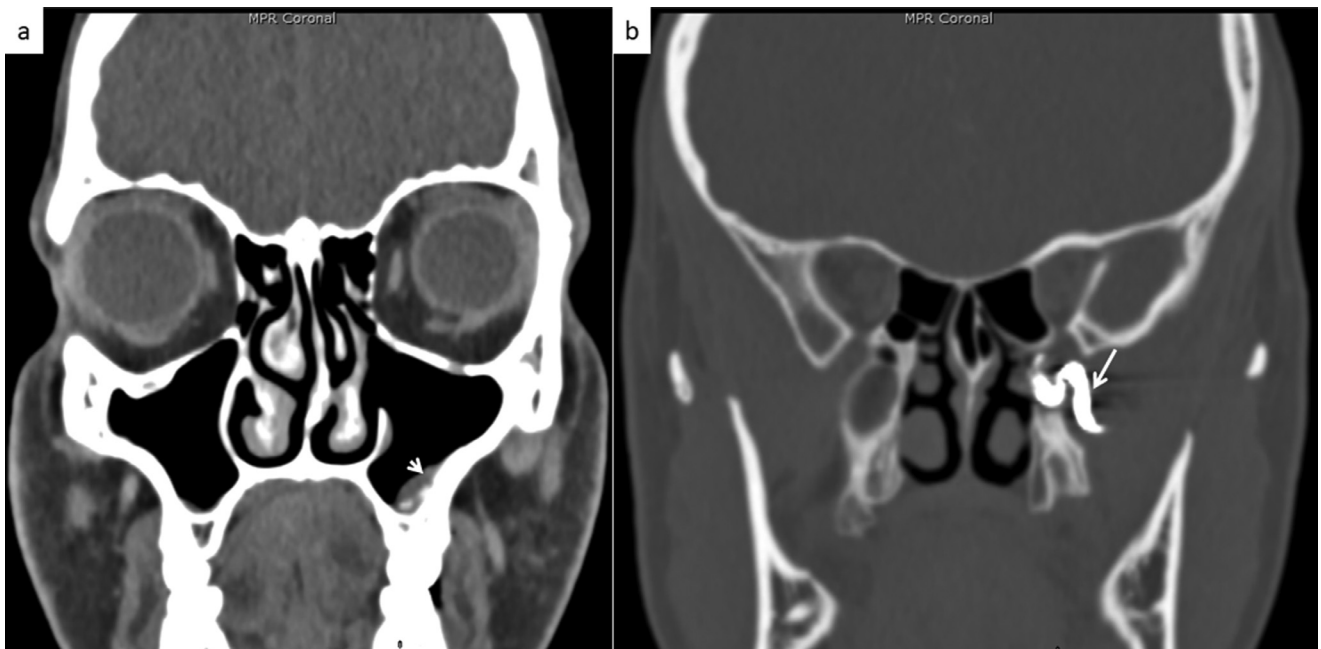


Fig. 5 – Postoperative CT scan in coronal reconstruction showing the total disappearance of the left sinus intramaxillary vascular lesion with mucosal thickening on the parenchymal window (arrowhead), and the embolization material centered on the left sphenopalatine artery on the bone window (arrow).

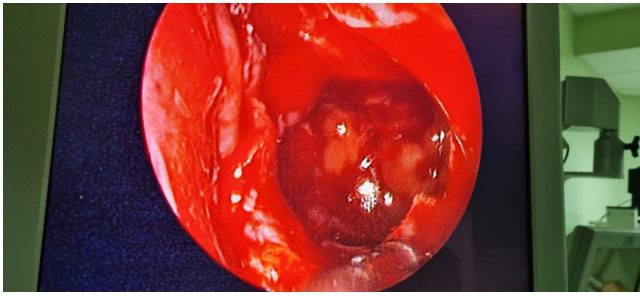


Fig. 6 – Intraoperative endoscopy image of hemangioma of the maxillary sinus.

The major clinical sign is epistaxis. Endoscopy reveals a regular red mass, rounded with a normal sinus mucosa. Facial CT shows a lesion of tissue density, enhancing after injection of contrast agents products, with areas that may be partially masked by hemorrhagic rearrangements, or necrotic alterations. It allows to determine the location, the extension of the tumor, and the bone involvement. On MRI, the lesion presents a heterogeneous signal on the different sequences, with a heterogeneous hypersignal T2 related to hemoglobin degradation products, and intratumoral vascular thrombosis. The lesion is surrounded by a T2 hyposignal border secondary to hemosiderin deposits at the periphery of the mass. The presence of hypointense foci is in favor of phleboliths, more characteristic of cavernous hemangiomas. MRI allows a better assessment of the lesion limits, of the tumor extension to the deep spaces of the face, and to differentiate between sinus invasion, which is seen in malignant tumors, and simple retention [2]. The lesion appears irregular on CT scan and well limited on MRI, well differentiated from the adjacent inflammatory mucosal thickening.

Arteriography specifies the arteries supplying the tumor and allows an embolization in order to facilitate surgery, by reducing intraoperative blood loss. The nourishing arteries often originate from the internal maxillary, ascending pharyngeal and facial arteries [2].

Since clinical signs are similar to other pathologies such as organized hematoma, papilloma, schwannoma, carcinoma, sarcoma, lymphoma, hemangiopericytoma, and paraganglioma. Preoperative diagnosis is necessary, in order to plan noninvasive surgery in case of benign lesion [4]. Among these differential diagnoses, organized hematoma is the most difficult lesion to differentiate from a hemangioma. It develops from the accumulation of blood in the maxillary sinus secondary to various etiologies (trauma, surgery, predisposition to bleeding, hemorrhagic lesion, ruptured aneurysm of an arterial branch, etc). The lack of ventilation and drainage of the maxillary sinus helps to form a fibrous capsule that prevents resorption of the hematoma. Fibrosis with neovascularization, which causes repeated bleeding, leads to an increase in the size of the lesion, causing scalloping on the adjacent bone. The only distinguishing feature is the often patchy, nodular or irregular papillary enhancement within the lesion [5].

In malignant tumors there is a frank bone destruction instead of erosion or simple scalloping, associated with an inva-



Fig. 7 – Resected specimen.

sion of adjacent structures. In case of doubt about the diagnosis, an extemporaneous anatomic-pathological test is recommended to eliminate a malignant tumor that requires more invasive surgery.

Treatment of sinus hemangiomas is a large exeresis by endoscopic approach for small tumors, preceded by preoperative embolization.

Conclusion

Facial sinus hemangioma is a rare benign vascular tumor, which can be locally aggressive. An accurate preoperative diagnosis is necessary to plan the surgical procedure. Imaging shows a heterogeneous lesion, enhanced after contrast with sometimes erosions and scalloping on bone without invasion of adjacent structures. MRI specifies the limited contours of the lesion, with a peripheral T2 hyposignal border, and an obvious heterogeneous enhancement.

Patient consent

The patient has given his consent for the publication of this data.

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