

Cavernous Hemangioma Involving the Maxilla: A Case Report

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Abstract

Background: Intraosseous vascular lesions are rare, they seldom occur in facial skeleton and maxilla is one of the least reported sites of involvement.

Case Presentation: We report a 45-year-old woman with cavernous hemangioma of maxillary bone involving inferior turbinate. It showed osteolytic expansile nature in imaging. The mass was approached via facial degloving technique. Histologic evaluation revealed vascular lesion in favor of cavernous hemangioma.

Conclusion: Intraosseous vascular anomalies are one of the diagnoses of facial skeletal masses not routinely occurring to the mind especially in adults due to their rarity and various presentations. They can mimic other pathologies with regard to symptoms and imaging characteristics. They also should be differentiated from malignancies, though frequently the definite diagnosis will be uncertain until proven histologically.

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Background

Vascular anomalies represent a broad spectrum of disorders. The International Society for the Study of Vascular Anomalies (ISSVA) classification has three levels of division for vascular anomalies. According to this classification, vascular tumors are differentiated from malformation and vascular malformations are categorized as slow and high-flow. Finally, these vascular lesions are defined by their components as arterial, venous, lymphatic, or mixed. According to the ISSVA classification; cavernous hemangioma should be considered slow-flow vascular malformations (1).

Intraosseous vascular anomalies account for less than 1% of all bony tumors. They are usually found in the vertebral column and skull bones. Involvement of the facial bones occurs infrequently (2).

Here, we describe a case of cavernous hemangioma involving the maxillary bone.

Case Presentation

A 45-year-old woman presented with a 1 year history of right facial swelling. She mentioned a slow growing facial mass on right perinasal side without pain which showed a small increase in size during this time period. She denied any history of facial trauma or previous surgery.

She had mild right nasal obstruction but the history was negative for epiphora, blurred vision, tooth pain or previous sinonasal symptoms.

On examination there was a hard fixed 5×4cm mass on medial side of right cheek. The overlying skin did not exhibit any color abnormality or adhesion to the mass.

Nasal endoscopic evaluation showed a swelling in right middle meatus with intact mucosa. In intraoral examination there was no palatal bulging or teeth loosening.

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Cross sectional imaging with computed tomography (CT) revealed an osteolytic expansile lesion with fine trabeculae on right nasal side wall (Figure 1).



Figure 1. Computed tomography (CT) scan revealed an expansile bony lesion.

The patient underwent surgical excision of the mass. Under general anesthesia, exposure was made using facial degloving approach. The flap was elevated easily without any evidence of adhesion to the mass. The mass was excised with a 3 mm margin of surrounding bone using drilling and osteotomy.

The mass had occupied the whole thickness of ascending process of maxilla extending into the middle meatal area. Anterior part of inferior turbinate was involved and excised with the mass as a whole (Figure 2). We did not encounter any brisk bleeding during the procedure. The specimen was sent for histologic evaluation which reported a cavernous hemangioma.

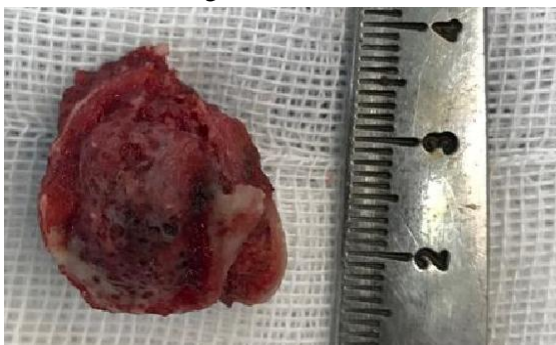


Figure 2. The specimen excised with anterior part of inferior turbinate.

Discussion

Intraosseous vascular anomalies are uncommon and maxillary involvement is a rare presentation reported in articles. Sáenz

and colleagues looked at about 100 cases of intraosseous cavernous hemangiomas published between 1845 and 2016 (3), none of them involved maxillary bone. Our literature review led us to 2 cases of maxillary bone involvement (4, 5)

Intraosseous cavernous hemangiomas present as painless slow growing masses which can lead, based on site of involvement, to dental loosening and tilting (6), neurologic symptoms (7) facial deformity (8-10), visual disturbance (11), nasal obstruction (12).

Early diagnosis and appropriate management of intraosseous hemangiomas is helpful in preventing the associated morbidities such as significant skeletal deformities and permanent facial defects. Although there is not a pathognomonic pattern, imaging findings are useful in showing features suggestive of the diagnosis. Borders of these lesions may be well-defined and corticated, or ill-defined, simulating a malignant tumor such as in our case (13, 14). It often shows an expansile process with a high density amorphous mass and multiple interposed trabeculae (15). Honeycomb, soap bubble and tennis racket appearances also have been reported in intraosseous hemangiomas (16).

Sunray pattern is a rare radiologic manifestation of cavernous hemangiomas (14) as seen in our case which led us to the possibility of osteosarcoma as one of our initial diagnoses.

Other differential diagnoses based on imaging findings, included osteoma, fibrous dysplasia, langerhans cell histiocytosis (15).

Histologically, the intraosseous cavernous hemangioma is composed of large thin-walled vessels and sinusoids lined with a single layer of flattened endothelium (4).

Osseous cavernous hemangiomas do not show spontaneous involution, so they need intervention to prevent their complications. Surgical excision is the treatment of choice, the possibility of recurrence is avoided by including a margin of safety although other

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options such as radiotherapy and embolization have been proposed (17, 18) In cases of full-thickness excision reconstruction options should be considered. Due to uncertainty about definite diagnosis, we decided to postpone probable reconstruction until histologic confirmation of disease. At 3-month follow up there was not significant facial deformity, also the patient did not complaint of epiphora and facial hypoesthesia.

Conclusion

Intraosseous vascular anomalies are one of the diagnoses of facial skeletal masses not routinely occurring to the mind especially in adults due to their rarity and various presentations. They can mimic other pathologies with regard to symptoms and imaging characteristics. They also should be differentiated from malignancies, though frequently the definite diagnosis will be uncertain until proven histologically.

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Patient Consent

Obtained.

Conflict of Interest

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