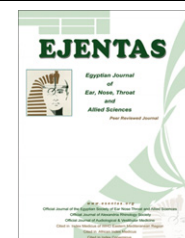




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Large central osteoma of maxillary sinus: A case report

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Abstract Osteoma is a benign osteogenic tumor arising from the proliferation of compact or cancellous bone. Osteomas of the craniofacial region are common in the nose and paranasal sinus particularly in the frontoethmoid region. However osteoma of the maxillary antrum is very rare and its presentation is different from the commoner frontoethmoid osteomas. We present here a rare case of large osteoma of the maxillary antrum in a 12-year-old child and also describe the difficulty faced in removing the tumor endoscopically. The tumor was removed using combined approach.

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1. Introduction

Osteoma is a benign fibro-osseous tumor arising from the proliferation of cancellous or compact bone.¹ Osteomas are commonly seen in the cranio-facial skeletal structures, especially in the nasal and paranasal regions. These are frequently seen in second and third decades of life, with male/female ratio of about 2:1.³ We report a case of large maxillary osteoma in 12-year-old female child.

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Osteoma is the most common benign fibro-osseous lesion that is present in the paranasal sinuses. It is situated in fronto-ethmoidal region in 95% of cases, involving the frontal sinus in 60–70% and the ethmoid in 20–30%, while it is rarely found in the nasal cavities, with the exception of the sphenoid sinus.⁷ The entity is particularly uncommon in the maxillary sinus, accounting for only 5% of cases.

Nasal and paranasal osteomas are usually asymptomatic slow growing tumors which can produce swelling and asymmetry of face. Ophthalmological and meningo-encephalic symptoms can rarely be associated with these tumors, especially the fronto-ethmoidal type.⁴ Maxillary sinus osteomas are usually asymptomatic which are diagnosed accidentally and require no treatment.

Surgical treatment is reserved for patients with infection or complication associated with compression to the neighboring structures. Surgical approach may vary from external approach to endoscopic approach.

The present report deals with the osteoma of the maxillary sinus in 12-year-old girl child which is exceedingly uncommon. The main clinical and therapeutic data regarding this tumor are reviewed and the difficulty faced in

removing the tumor solely by endoscopic approach is highlighted.

2. Case report

A 12-year-old girl presented to the outpatient Department of Otolaryngology with slowly progressive forward and upward displacement of the left eye over a five-month period. She also complained of double vision in the downward and left lateral gaze. She also had diffuse swelling of left malar region for 1 month. There was no history of recurrent rhinorrhoea, watering of eyes or nasal bleed. There was no history of notable facial trauma or previous nasal surgery and her medical condition was irrelevant.

Examination of eye disclosed a visual acuity of 6/6 in the right eye and 6/5 in the left eye. There was non-axial proptosis, with upward and forward displacement of the left eye. Hertel exophthalmometry revealed 8 mm of left eye proptosis. The extraocular movement was restricted in the downward and left lateral gaze. Pupils were normal size and normal reacting to light. No orbital masses could be palpated. Anterior and posterior rhinoscopy was normal. There was diffuse 3 × 3 cm bony hard swelling involving upper left malar region.

Computed tomography (CT) of nose and PNS showed a large, well circumscribed densely calcified lesion filling the left maxillary sinus in its upper part and extending upward into the left orbit (inferiomedial aspect) and medially involving the ethmoid sinus. It was reaching posteriorly till the orbital apex with proptosis of left eye (Fig. 1).

On the basis of clinical and CT findings diagnosis of osteoma of maxillary sinus was entertained. The plan was to excise the maxillary sinus osteoma endoscopically.

The patient was operated under general anesthesia. Using 0° endoscope into the nasal endoscope inferior turbinate was

removed. Uncinectomy was done and maxillary ostium was widened. There was dense whitish bony hard mass seen hanging from the roof of maxillary sinus. Using cutting burr drilling of the mass was started. The medial attachment of the tumor was made free. It was difficult to approach the posterior and lateral extent of tumor by endoscopic approach. So it was decided to remove the tumor by external approach. Weber-Fergusson incision was made. The anterior wall of maxilla was exposed. A window was made in the anterior wall of maxilla. There was a 4 × 5 × 3 cm bony hard mass filling upper part of left maxillary sinus going to the inferiomedial part of the orbit through a defect in the floor of the orbit (Fig. 3a). The osteoma was removed enmasse with help of endoscope. The defect in orbital floor was repaired with cartilage and prolene mesh.

CT scan performed one month after surgery showed no residual lesion (Fig. 2).

Histopathology of surgical specimen with hematoxylin and eosin staining showed relatively dense, mainly compact bone with sparse marrow tissue compatible with osteoma (Fig. 3b).

She last came for follow-up after four months of surgery. She was asymptomatic and had no ocular complaints and is doing fine.

3. Discussion

This case report describes a 12-year-old female with giant osteoma of maxillary sinus extending into the orbit. Osteomas in the maxillofacial region show a predilection for the mandible, especially the ramus and the inferior border below the molars.¹⁰ Furthermore, the paranasal sinus osteomas, with their reported incidence ranging from 0.01% to 0.43%, are generally found in frontal, ethmoidal and rarely in maxillary sinuses, in decreasing order of frequency.⁹ They are usually

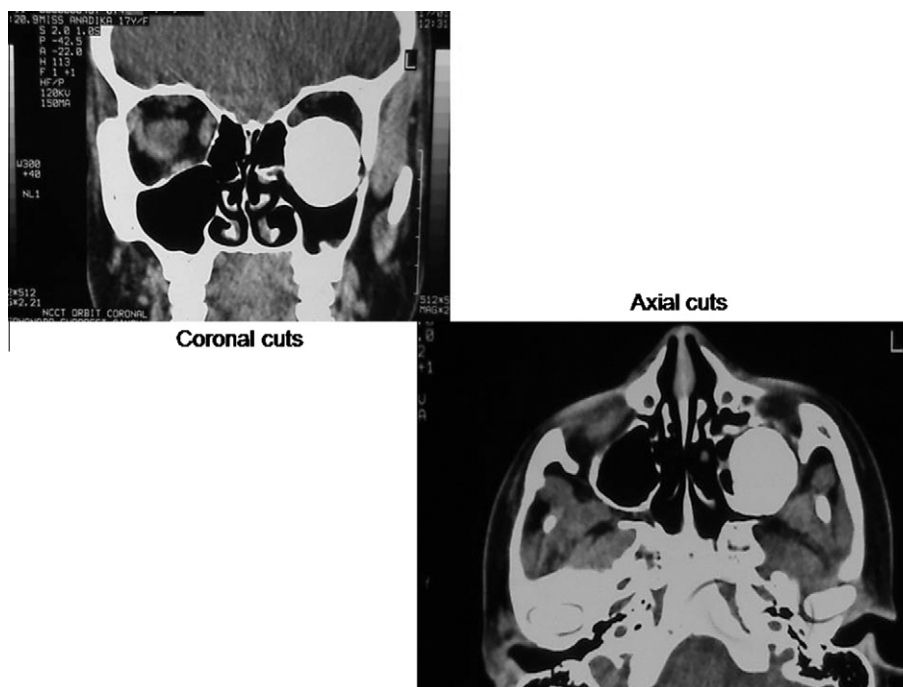


Figure 1 Computed tomography of nose and PNS (coronal and axial cuts) showing dense radio-opaque mass arising from the roof of maxillary sinus extending upwards into the orbit with breach in the inferior wall of orbit and also filling the maxillary antrum.



Figure 2 Post operative CT scan of nose and PNS (coronal and axial cuts) after 1 month showing complete removal of the mass.

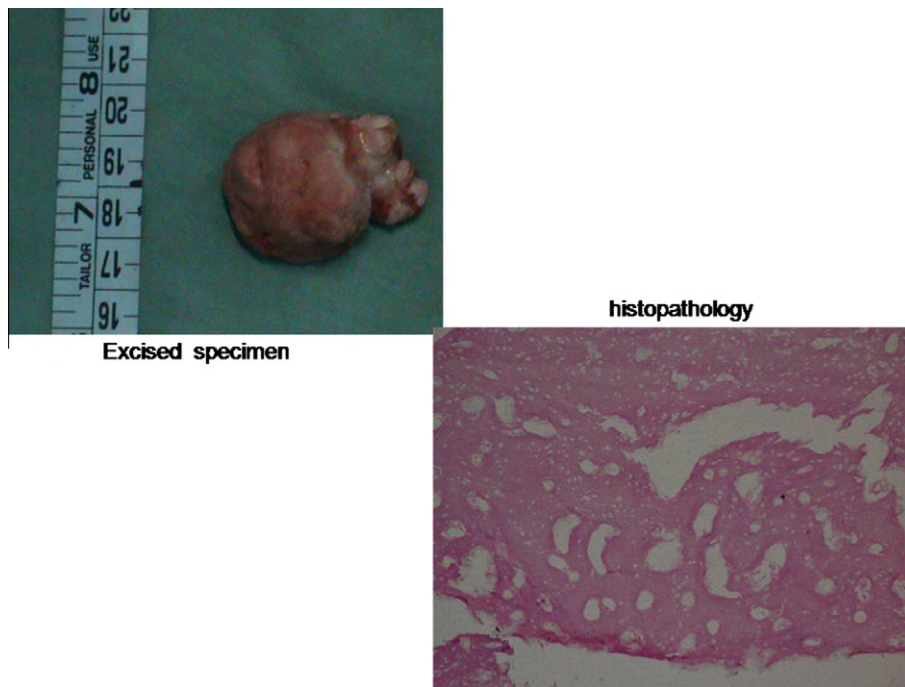


Figure 3a Excised surgical specimen removed in toto.

asymptomatic and are discovered as a coincidental radiological finding. From this point of view, the lesion in our case may be considered rare and atypical, not only for its unusual location, but also for its dimensional features.

The pathogenesis of these tumors still remains to be elucidated, but several theories have been advanced in this respect. Embryological theory suggests that these osteomas arise from

osseous proliferation due to the apposition of membranous and enchondral tissue forming close to the bony sutures, such as fronto-ethmoidal one.² Traumatic theory has been suggested in a number of papers, according to which the development of these tumors is due to traumatic events in the past (in 20% of cases), particularly in males and during puberty, when skeletal growth is at its peak.² Minor trauma followed by

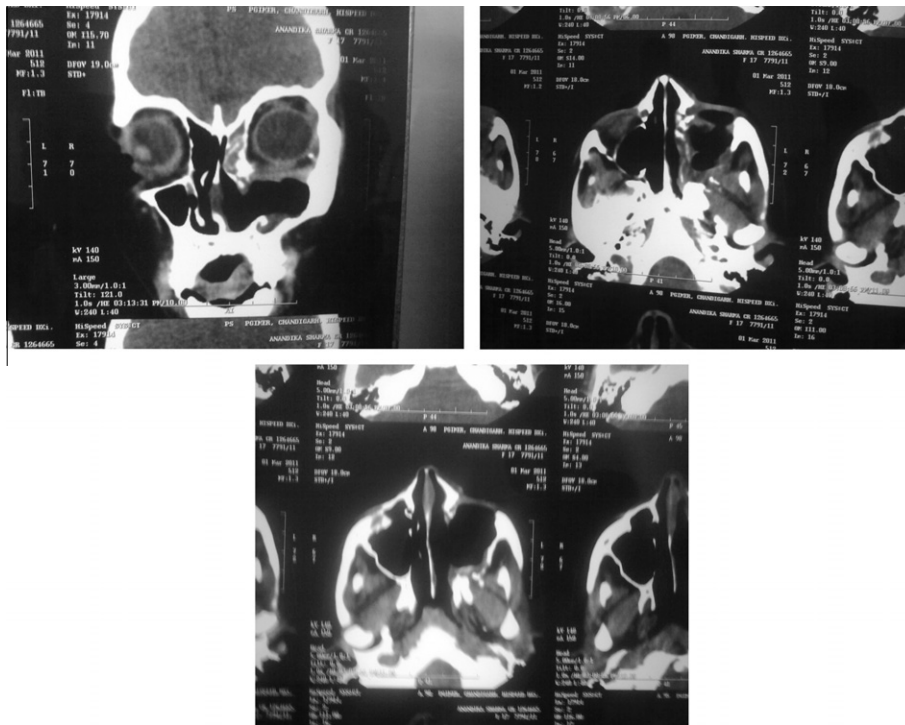


Figure 3b Photomicrograph of the mass showing a well outlined mass comprising of interconnected bony trabeculae and well developed vascular spaces in between (H&E, $\times 150$).

periosteal bleeding and edema formation has been suspected to trigger a reactive osteogenic process that initiates the abnormal development of bone structure. Inflammatory theory suggests a determinant role that may possibly be played by a previous inflammation of the sinuses, which would act as a stimulus for the proliferation of osteoblasts situated along the muco-periosteal junction of paranasal sinus; the calcification surrounded by inflamed tissue would subsequently give rise to the osteoma. The two latter theories, traumatic and inflammatory causes, have often claimed to be responsible for osteomas of maxillary sinus. However, our patient did not have any history suggestive of previous trauma or infection.

According to the clinical and radiological topography, three variants of osteoma can be distinguished: central, peripheral or extraskeletal. Central osteomas arise mainly from endosteum, whereas peripheral variants originate from the periosteum and the extra skeletal type resides within a muscle.⁸ The peripheral type quickly produces swelling, asymmetry and erosion of the surrounding structures.¹ Differential diagnosis between central and peripheral types often confuses the clinician. Intraoral clinical examination of our case revealed no bony specula or prominent mucosal swelling in the affected region, indicating that the lesion did not cause significant resorption of the neighboring bone. The results of CT scanning also supported this assumption as buccal and palatal bone cortices of the maxilla were found to be intact. Moreover, during the surgical intervention, the bony-hard mass was observed to be directly connected to the surrounding bone. These details have led us toward the diagnosis of a centrally localized osteoma of the maxilla.

Histologically, these are described as either eburneous, if they are composed of solid bone, or spongy, when they have gaps in the marrow of irregular structural arrangement. Both

types can be situated in maxillary sinus, but the eburneous form tends to be frontal while the spongy version is usually ethmoidal.

In the clinical setting, osteomas develop mainly during skeletal growth and are characterized by an independent development and slow progression related to peripheral subperiosteal bone apposition which creates a spreading, puffy shell. As it spreads gradually outward, the osteoma can produce marked deformities of the face, neuralgia and nasal and paranasal inflammatory processes, but if it develops mainly inside the sinus it may not be perceived for a long time. Diagnosis is usually made by chance, usually during X-ray examination of the skull (in 40% of cases) due to facial pain or headache, especially if the site of osteoma is fronto-ethmoidal or sphenoidal.²

Radiologically, the osteoma is seen with a more or less homogeneous radio-opaque lesion, attached to the sinus wall with a small stem. CT is considered to be the most suitable imaging modality for the diagnosis of osteoma. The use of CT scan with 3-D reconstruction makes it possible to achieve a better resolution and more precise localization.

It is important to rule out Gardeners syndrome in patients of osteoma. These patients may present severe gastrointestinal disorders with rectal bleeding and abdominal pain. The triad of colorectal polyposis, skeletal abnormalities and multiple impacted teeth is consistent with this syndrome.⁶

Paranasal sinus osteomas usually grow slowly. However, Koivunen et al.⁵ reported that the growth rate varies remarkably by pointing out that some osteomas could be in the same size after a long follow-up period, but the growth rate can also be as much as 6 mm/year without any specific factor. For that reason, they suggested that a paranasal sinus osteoma should

be removed if the lesion fills 50% of the volume of sinus or if it causes any symptoms.

Whether surgery is to be performed depends on the size of osteoma, any worsening of symptoms and the possible onset of infection and/or complications due to compression. Osteomas are usually removed either in toto or broken up and sucked out under direct control. For the maxillary antrum, the sub-labial gingivo-buccal (Caldwell-Luc) approach is convenient. Endoscopic techniques have been advocated in selected cases. There have been recent reports of use of the endoscopic nasal approach for the resection of ethmoidal and frontal osteomas. Removal can be difficult and incomplete, with possible recurrence of the tumor, when performed using endoscopic methods, especially in case of solid maxillary sinus tumors. Besides we found it difficult to reach the most posterior and lateral part of osteoma with endoscopic approach. So we advocate endoscopic assisted approach for the removal of maxillary osteomas.

Recurrence of paranasal sinus osteomas after surgical excision is extremely rare. There are no reports of malignant transformation of osteomas in the literature.

To conclude, although completely curable with adequate surgical treatment, osteomas of the paranasal sinuses still present a diagnostic challenge depending mostly on their late discovery and unpredictable behavior.

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