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Case Report

Surgical Management of Ossifying Fibroma in Maxilla: Report of Two Cases

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Abstract:

Ossifying fibroma is a rare benign osteogenic neoplasm arising from undifferentiated cells of the periodontal ligament. Ossifying fibroma have a well-defined border that differentiates it from fibrous dysplasia clinically, these tumors manifest as a round or ovoid, expansile, painless, slow-growing mass may displace the roots of adjacent teeth and also cause root resorption. They occur at second to fourth decade of life. Radiologically the lesion appears as a dense radiopaque mass surrounded by a thin, well-defined regular radiolucent rim. Patient underwent thorough history taking and complete face, ear, nose, and throat examination. Computed tomography maxilla, orthopantomogram, paranasal sinus reveals entire maxillary sinus involved in one case. Nasal septum deviated to the opposite side, airway reduced on the side of swelling seen in one case. Root resorption seen in two cases and missing teeth in seen in case 1. In our study in case 1, the tumor involved maxillary sinus, medial wall of the nose. The tumors were excised by Weber–Fergusson and in case 2 the tumor was excised by maxillary vestibular approach. Overall recurrence rates after resection is reported to range from 30 to 56%.

Key Words: Bone neoplasm, cemento-ossifying fibroma, fibro-osseous lesion, maxilla, ossifying fibroma, Weber–Fergusson approach

Introduction

Ossifying fibroma is an uncommon benign osteogenic neoplasm, and it is derived from the multipotential mesenchymal cells of the periodontal ligament.[1] Ossifying fibroma is a "well-circumscribed lesion, which consists of fibrous tissue, highly cellular in nature with varying amounts of calcified tissue, which gives the appearance as the bone, cementum or both.[2]

Clinically, these tumors are round or ovoid, expansile, painless, slow-growing mass that displace the roots of adjacent teeth, and it may cause root resorption. They occur in second to fourth decade of life with a definite female predilection and maxilla is affected less than mandible, most common site of occurrence is premolar - molar region of the mandible.[3] Su *et al.*[4] 70% cases reported mandible is the most common site of occurrence, with 43% located in the mandible posterior region, including the ramus area, followed by 22% located in the maxillary posterior region.

Although the precise pathogenesis is still unknown, but Wenig *et al.*[5] has suggested that trauma – induced stimulation may play a role. In our case study, we are reporting two cases of an unusual occurrence of ossifying fibroma in maxilla and its surgical management.

Case Report

Case 1

Clinical presentation

A 28-year-old female presented to our Oral and Maxillofacial Department with a painless, progressive swelling of the right face since 2 years. Extraoral examination demonstrated a moderately large left facial swelling. There was no facial anesthesia. Extraocular eye muscle movements were normal. A painless swelling in the right maxilla and approximately 4.45 cm × 5.06 cm in size. Clinically the lesion was firm to bony hard in consistency and located between the 11 and 17. Intraoral examination, expansion of the left maxillary alveolar process that is extending to the hard palate (Figure 1). There was minimal tenderness on palpation. The right maxillary molar and premolar teeth were loose. Cervical or submandibular lymph nodes are not palpable. Her medical history was insignificant. She had no other complaints.

Imaging

Computed tomography (CT) scan revealed a mixed appearance of radiolucency and radiodensity, and a well-defined osteolytic mass involving the right maxilla, maxillary sinus, alveolar bone and nasal cavity. There was destruction of the medial and antero-lateral walls, and the roof of the left maxillary sinus. The inferior orbital bone was partially involved (Figure 2a and b).

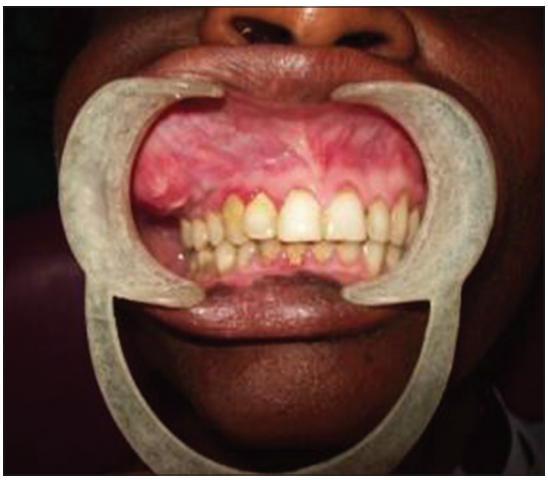

Figure 1: Clinical view (hard bony round swelling extending from right side anterior to posterior maxillary vestibule, 13-17 and buccal cortical plate expansion).

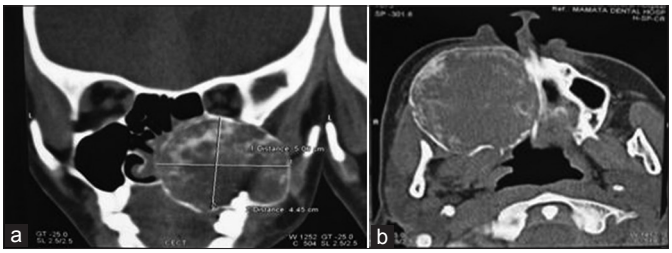

Figure 2: (a)Computed tomography coronal section (well defined mixed radio opaque - radiolucent lesion involved medially medial wall of the nose, superiorly infra orbital rim, laterally posterior wall of the maxillary sinus, inferiorly anterior wall of the maxillary sinus, size of the lesion 4.45 cm × 5.06 cm). (b) Computed tomography axial section (dense radio opaque round mass surrounded by thin, well defined radioopaque rim and complete obliteration of the maxillary sinus).

Management

After routine work-up and obtaining anesthetic fitness, the patient was taken to the operating theater. Under general anesthesia, with all aseptic conditions, the tumor was exposed and a complete excision was performed through a Weber–Ferguson approach. A right total maxillectomy was made, and the orbital floor invaded by the lesion was also removed (Figure 3a and b). Intra-operatively, the tumor was found to be well-encapsulated with a cleavage plane to allow it to be shelled out from its surrounding structures. The orbital and the nasal floors were preserved and maintained. The palatal mucoperiosteum was preserved, and the tumor was removed en masse in one piece with no perforations and layer by layer closure was done by vicryil and 3-0 BB silk sutures are placed. Post-operative recovery was uneventful. Patient remains asymptomatic for the past 8 months with minimal facial disfigurement and minimal visible facial scar. Clinical and radiographic evaluation of the upper right quadrant shows no change of the lesion, which we are planning to follow-up

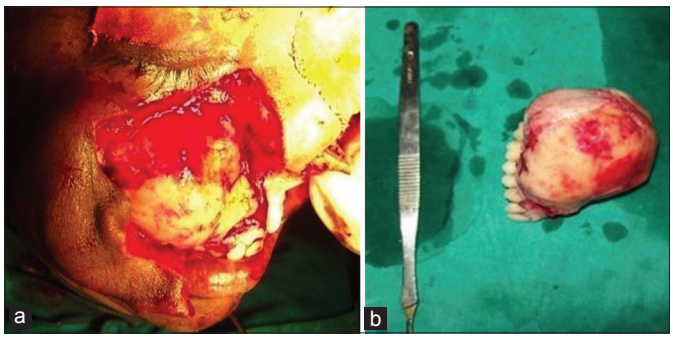

Figure 3: (a) Intra-operative view (excision of the lesion through Weber–Fergguson approach). (b) Resected encapsulated mass (complete shell out the encapsulated mass from its surrounding structures).

periodically. Review additionally, the orbital floor invaded by the lesion was also removed. The patient had an excellent outcome, with very good prognosis.

Case 2

Clinical presentation

An 18-year-old young male presented with a complaint of progressive swelling of the left maxilla obstructing his right nostril and causing disfigurement of the left upper lip. According to the patient, the mass had been slowly expanding since 7 months. Though, the pain was not a presenting feature. There was no regional lymphadenopathy. The patient's general health was good with no co-morbid conditions. There was no associated family history of similar type of tumors. Clinical examination revealed a well-circumscribed, slow-growing lesion causing massive bony expansion. The mass was firm to hard in consistency. The mass caused a facial disfigurement with the swelling extending from the corner of the mouth region. Intra-orally, the buccal vestibule was completely obliterated with the mass extending from the left canine to first premolar. Left canine and first premolar teeth are missing. Palatally, the soft tissue was healthy with minimal expansion of the palatal alveolus (Figure 4). The vault of the palate was not involved, and the midline was not violated.

Imaging

CT scan confirmed the extent of the mass. It also established that in spite of the massive expansion, there were no noticeable perforations and showed that the border of the lesion was completely well-defined and it was encroached to the floor and lateral wall of the sinus with cotton-wool appearance (Figure 5).

Management

After routine work-up and obtaining anesthetic fitness, the patient was taken to the operating theater. Under general anesthesia, the tumor was surgically removed through a maxillary vestibular approach. Intra-operatively, the tumor was

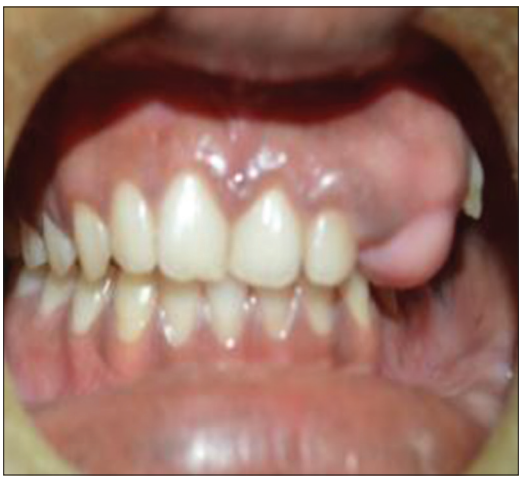

Figure 4: Clinical view (ovoid bony hard swelling in the left anterior maxilla. Missing teeth in relation to 23 and 24. Buccal cortical plate expansion).

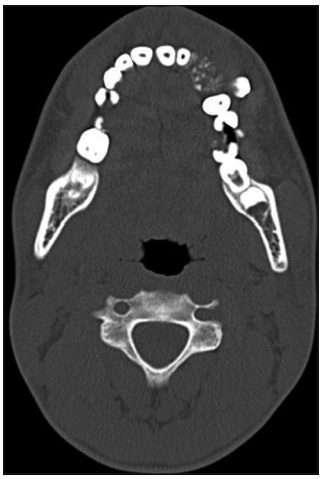

Figure 5: Computed tomography axial section (encapsulated ovoid radiolucent lesion. Left maxillary canine and premolar teeth are missing).

found to be well-encapsulated with a cleavage plane to allow it to be shelled out from its surrounding structures. The orbital and the nasal floors were preserved and maintained. The palatal mucoperiosteum was preserved, and the tumor was removed en masse in one piece with no perforations. Surgical cavity was debrided completely (Figure 6a and b). Post-operative recovery was uneventful. Patient remains asymptomatic for the past 8 months with minimal facial disfigurement and no visible facial scar. Clinical and radiographic evaluation of the upper left quadrant shows no change of the lesion, which we are planning to follow-up periodically.

Discussion

Multiple familial ossifying fibromas was first stipulated by Yih *et al.*[6] Ossifying fibroma occurs in the second to fourth decades of life (50%), with a female predilection (100%). In our cases, age ranged between 18 and 60 years.[2] Although Krausen *et al.*[7] reported no particular sex predilection and a common

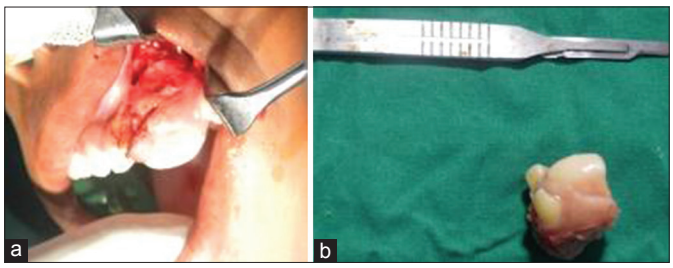

Figure 6: (a) Intra-operative view (excision of the lesion through maxillary vestibular approach). (b) Resected encapsulated mass (complete shell out ovoid encapsulated mass from its surrounding structures).

occurrence in the premolar-molar area of the mandible and above the mandibular canal, it has also been reported in the orbital and petro mastoid regions, and the maxillary, ethmoidal, frontal and sphenoidal sinuses too. Ossifying fibromas of the maxillary sinus are unusual tumors. If it occurs in the maxilla, it is most likely to be found in the canine fossa and the zygomatic arch area. In our study, the lesion was involved in anterior to the posterior region of the maxilla in one case and canine fossa region of the maxilla in one case.

Clinically, massive bone expansion of buccal and or lingual cortical plates is the most common clinical sings of ossifying fibroma.[8] Due to the slow growth, the overlying mucosa or skin and the cortical plates of the bone are invariably intact. Ossifying fibromas are usually asymptomatic in nature until noticeable swelling is observed and facial asymmetry.[9] In our cases, the patients presented with symptomatic swelling, pain and nasal block that persisted for a long duration in case 1. In case 2, patient had pain swelling since 7 months.

On the contrary, Barberi *et al.*[10] (2003) reported the lesion without scelorotic rim (40%), defined lesion with sclerotic rim (45%), and lesion with ill-defined border (15%). In our study, both Cases satisfy the findings of Waldron *et al.* and Barberi *et al.*[10] Radiologically, the lesion presents different stages of development with a centrifugal growth pattern. Sometimes the lesions may grow massively and invades in all direction and gives the appearance as round tumor masses. A thin fibrous capsule demarcates the lesion from the adjoining normal bone.[11] In the present study, tooth displacement was seen in one case as tilting, but root resorption was seen in two cases and missing teeth in one case.

Histopathological examination shows a pattern with small irregular bony trabeculae that are surrounded by osteoblasts. The lesion consists the collagen stroma, containing variable numbers of uniform spindle on stellate cells. Woven bone patterns seen in the early tumors when assessed under polarized light, and osteoblastic rimming is minimal in the mature lesion and the irregular trabeculae.[11]

Treatments of choice for small, well-circumscribed lesions are enucleation. Baumann, Zimmermann *et al*. 2005; reported, despite its benign features, large lesions can be locally infiltrated to adjacent structures, causing significant morbidity, and fatal consequences may be induced by intracranial extension. Newman *et al*. 2009, based on anatomic location and tumor size will guide the surgical approach than by histologic subtype shields.[12] Liu *et al*.[11] identified that recurrence rate is unpredictable, from 6 months to 7 years.

In our study, in the case 1 the tumor involved antero-postero lateral walls of the maxillary sinus, medial wall of the nose. Pre-operative evaluation of all the patients was done by the general physician and anesthesiologist. All the patients were fit for general anesthetic procedure under ASA Grade I (healthy patients with no systemic disorder).

Under general anesthesia through nasotracheal intubation was done and preparation of the surgical site with betadine solution is carried out. Then the Weber–Fergusson's incision marking on the face, this incision runs vertically through the center of the upper lip from the red margin to the base of the columella. An equally good result can be obtained by following the philtral prominences. At the base of the columella, the incision turns horizontally running in the angle between the nose and the lip, the cheek around the alar base. Then, the incision turns up along the side of the nose almost to the inner canthus. Before actually putting the incision, its line should be drawn with Bonney's blue and matching points tattooed for subsequent suturing. From the inner canthal area, the classical Fergusson's incision runs laterally across the lower eyelid at a distance from the lid margin. The placing of the incision in this line is recognized to result in intractable lymphedema of the eyelid. To avoid this modified version is used, which runs parallel to and 2-3 mm from the lid margin. The skin of the eyelid is elevated as a part of the cheek flap leaving the greater part of orbicularis, the tarsal plate, and the conjunctiva *in-situ*. The upper lid is divided into full thickness, and the incision is continued backwards along the upper buccal sulcus to the maxillary tuberosity. The cheek flap is then elevated off the underlying maxilla. Thus, the Fergusson's incision in its classic and modified form gives a wide exposure. If orbital extension operation is carried out along with maxillectomy, the incision can be further extended to encircle the lid margins.[13]

In case 2, the tumor was surgically removed through a maxillary vestibular approach. Chauvet *et al*. 2009, 30-56% is overall recurrence rate due to the infiltrative nature of the tumor borders causes incomplete excision.

Conclusion

Distinguishing between ossifying fibroma and other fibrous osseous lesions is the primary diagnostic challenge. In our case reports describing the clinical and radiological features and finally confirmed the diagnosis of ossifying fibroma. Enucleation and curettage are the treatment of choice in small, well-circumscribed tumor. In very large lesions, the tumor infiltrate into the surrounding structures, anatomic location and tumor size will guide the surgical approach than by histologic subtype shields. In our cases, case 1 complete maxillary sinus was obliterated and medial wall of the nasal wall also involved, Weber–Ferguson approach was used and case 2 the tumor was small in size, well-circumscribed border, maxillary vestibular approach was used.

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