Case Report

Extensive ossifying fibroma of jaw

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ABSTRACT

Ossifying fibroma of bone is a central neoplasm of bone and it is more common in young adult with marked predilection for mandible and also it is more common in female. Lesion is generally asymptomatic until growth produces a noticeable swelling and mild deformity. It presents an extremely variable roentographic appearance depending upon the stage of development. This lesion is composed basically of many delicate interlacing collagen fibers, seldom arranged in discrete bundle, interspersed by large numbers of active, proliferating fibroblasts. The lesion should be excised conservatively. We present a case of huge ossifying fibroma arising from maxilla.

Key words: Fibro-osseous, maxilla, ossifying

Introduction

Cemento ossifying fibroma (COF) is a benign osseous tumor, which is very closely related to other lesions such as fibrous dysplasia, cementifying periapical dysplasia or cemento-osseous florid dysplasia.[1] It is a bony tumor of the possible odontogenic origin. It is believed to arise from the cells of the periodontal ligament.[2] It has multipotential cells capable of forming cementum, lamellar bone and fibrous tissue. Under pathological conditions neoplasms may contain any or all of the components.[3] More aggressive lesions usually involve the maxillary antrum where extensive growth is unimpeded by anatomic obstacles.[3] Tumor manifest as slow-growing, asymptomatic, intraosseous masses, most frequent in females between 35 and 40.[4,5] COFs of the mandible are common, but COFs of the maxillary sinus are rare and few have been reported in the literature.[4] Surgical excision is the treatment of choice 4. Recurrence is variable.[6]

Case Report

The present case report is about a 45-year-old female patient who reported to our department; in with the chief complaint of huge swelling on the right side of the face, which was gradually increasing in size since last 6 year. He had a history of tobacco chewing since last 15 years. On examination swelling was 9.0 cm × 10.0 cm in size at right maxillary region displacing the right alae of nose, columella and septum on the left side. Superiorly the swelling displaced the lower eyelid compressing the eyeball giving the sun-set appearance of the eye. Inferiorly the swelling distorted the normal appearance of upper lip. Laterally the swelling has involved zygomatic region and up to the preauricular region [Figure 1]. Superficial skin was normal and non-adherent to lesion, but stretched with prominent vascular margin. On palpation the swelling was non-tender, non-compressible and non-reducible. Regional lymph nodes were not palpable.

Intra orally swelling was extending from left central incisor to right second premolar with palatal extension up to mid palatal line obliterating the buccal sulcus. The swelling has displaced all teeth from right central incisor to second premolar and with normal appearing mucosa but stretched with multiple erythematic patches. Mouth opening was normal but restricted lateral movement of mandible was noted. Teeth involved in the lesion were Grade I mobile and non-tender. Hematological, biochemical and other investigations were within the normal limits.

Incisional biopsy was done under local anesthesia. Histological examination of specimen revealed many delicate interlacing collagen fibers, seldom arranged in discrete bundles, interspersed by a large number of active proliferating fibroblasts with occasional presence

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DOI: 10.4103/0975-5950.154843

of cementum like droplets. On the basis of these, the diagnosis of COF was made.

Computed tomography scan revealed a 8.0 cm × 8.5 cm × 10 cm expansile lesion arising from the right half of the hard palate and maxillary alveolar arch. The lesion showed mildly enhancing high attenuation material with foci of calcification dispersed in it. Multiple calcifications of size and shape were observed. The lesion showed well defined anterior margin and irregular and eroded posterior and lateral margin. Superiorly the lesion was eroding the right inferior orbital wall. However, no intra-orbital extension was seen. Posterosuperiorly the lesion had displaced the maxillary sinus, which appeared collapsed. Medially the lesion was involving the right nasal fossa resulting in deviation of the nasal septum towards the left side. It is also involving anterior ethmoidal air cells. However, it was not crossing the midline. Laterally the lesion is involving inferior and lateral part of right zygomatic arch. The soft-tissue overlying the lesion all over is thinned by the lesion [Figures 2 and 3].

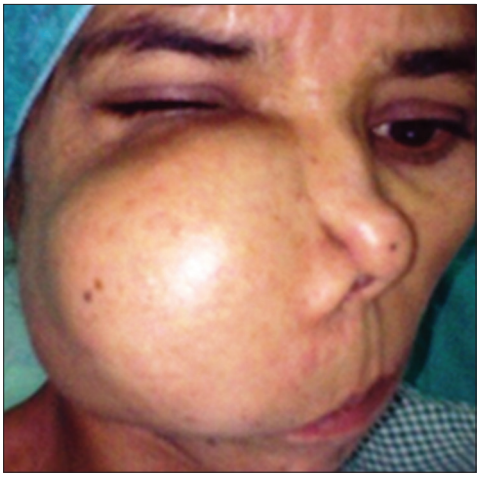

Figure 1: Extra oral presentation of lesion

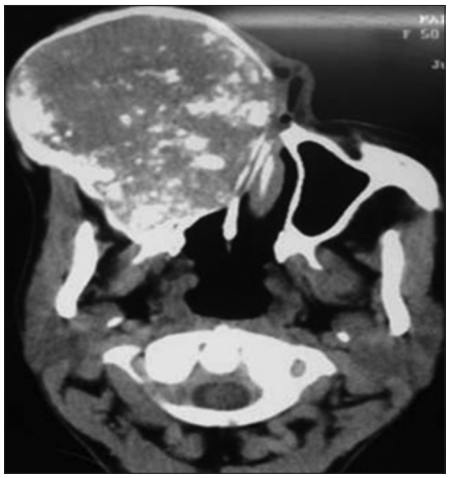

Figure 3: Computed tomography scan demonstrate extent of lesion

As per histological findings patients was planned for excision of the lesion. The lesion was excised until healthy margin of bone were achieved, through extra oral Weber-Ferguson approach under general anesthesia. Per operatively the lesion was encapsulated. It is removed with minimal bleeding and minimal injury to vital structure [Figure 4]. Excised specimen [Figure 5] was submitted for histopathological examination, which confirms the diagnosis of ossifying fibroma [Figure 6].

Post-operatively patient had mild post-operative hematoma of superficial skin on the right side of the face. The level of the right eye was restored to normal which was elevated pre-operatively due to massive size of the lesion. Anatomic structure such as alae of nose, columella, septum and upper lip was also restored to normal posture.

The above mentioned case is presented hereby because of its huge size causing facial asymmetry which is very rare. It was a case in which the lesion rarely presents in the maxilla with a weight of the lesional mass was 350 g.

Discussion

Fibro-osseous lesions of craniofacial skeleton are rare and believed to be the result of replacement of

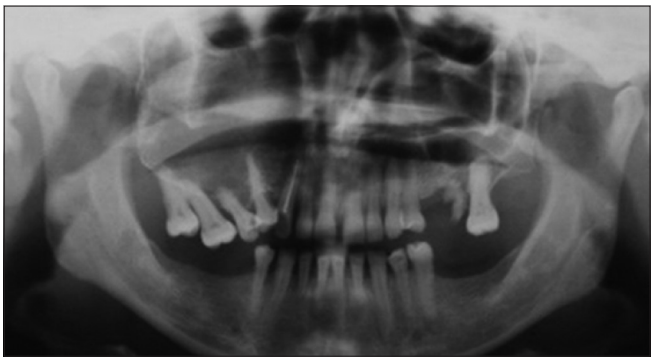

Figure 2: Orthopantomogram demonstrate extent of lesion

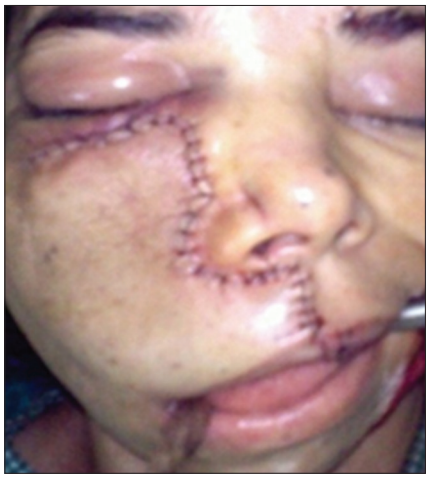

Figure 4: Post-operative profile view

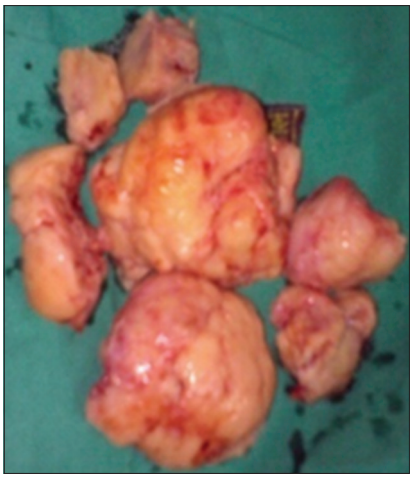

Figure 5: Excised specimen

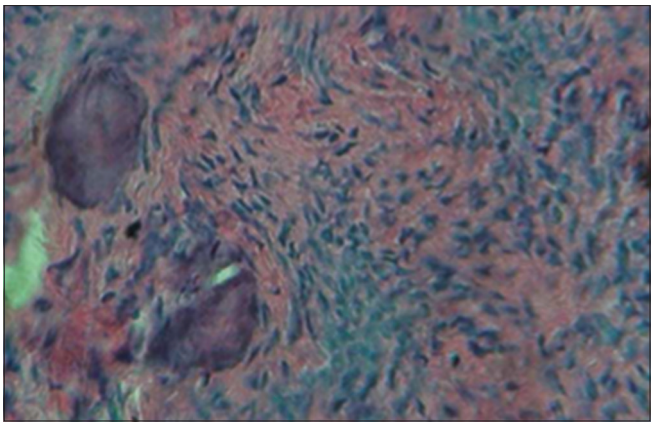

Figure 6: Histomicrograph of specimen

normal bony architecture by fibrous tissue, which may, mineralize in various forms such as woven, lamellae bone, or cementum and include a broad spectrum of distinct entities with different clinical presentations and microscopic appearance.[7]

The calcifications are extremely variable in various stages of bone and cementum deposition. Histologic differentiation between osteoid and cementum is difficult. In some cases, most of the calcified fragments are immature cementum; these tumors are named central cementifying fibroma. In other cases, the calcified fragments are osteoid, these tumors are named central ossifying fibroma. Central cementifying fibroma and central ossifying fibroma arise from the same progenitor cell but produce variable amounts of bone and cementum within any one lesion. The hybrid term central COF may be used to designate the presence of both types of tissue within the same lesion because of the difficulty in being able to distinguish reliably immature bone from immature cementum and because of the presence of both of these substances in many of the lesions. Thus, central COF is the most accurate term; it can be interchanged with either central ossifying fibroma or central cementifying fibroma. There is no apparent clinical or radiologic difference between the central cementifying fibroma or central ossifying fibroma.[8]

The mean age of patients at the time of diagnosis is 34 years (range, 16-62 years).[4] There is a definite female predilection. The mandible is involved more often than the maxilla, especially the premolar and molar regions.[5,6,9] Nearly 93% of COFs are found in the mandible.[4] Predilection for the mandible has been demonstrated in various studies.[5,6,9] In the 64 cases of COF reported by Eversole *et al*.,[5] the lesions were found most frequently in the molar region (52%), followed by the premolar (25%), incisor (12%) and cuspid (11%) regions. Presence of lesion was also reported in maxilla.[4]

Swelling displaced alae of nose, columella and septum towards the left side, lower eyelid displaced superiorly giving rise sun-set appearance due to aggressive growth which is consistent with features described in the literature. Bone swelling or expansion at the buccal and/or lingual cortical plates is the most frequent clinical sign of COF (96%,).[4] Similar results have also been noted in previous studies.[5,6,9] 19%, caused displacement of the roots of adjacent teeth.[2] Root displacement has also been demonstrated in 17% and 18% of COFs reported by Eversole *et al*.[5] and Sciubba and Younai[9] respectively.

Fibrous dysplasia and COF are clinically and radiologically distinct disease entities that nevertheless are not always histologically distinguishable.

Differential diagnosis of COF depends on the radiographic features of the lesion. COF with a completely radiolucent lesion may be misdiagnosed as early stage of cemento-osseous dysplasia, odontogenic cyst, Periapical granuloma, traumatic bone cyst, ameloblastoma, or central giant cell granuloma. COF with mixed radiographic features might be given a nonspecific diagnosis of fibroosseous lesion, or misdiagnosed as a calcifying odontogenic cyst (Gorlin cyst) or an adenomatoid odontogenic tumor.[4] Other differential diagnoses of COF with mixed radiographic features may include rarefying and condensing osteitis, intermediate stage of cemento-osseous dysplasia, fibrous dysplasia, calcifying epithelial odontogenic tumor (Pindborg tumor), or odontogenic fibroma. Furthermore, COF with completely radio-opaque radiographic features may be misdiagnosed as retained root, odontoma, idiopathic osteosclerosis, condensing osteitis, late stage of cemento-osseous dysplasia, or osteoblastoma. COF with a very large size may be misdiagnosed as an osteogenic sarcoma.[4] Early lesions may be radiolucent as they mature, they become a mixed radiolucent and radio-opaque lesion and finally become radio-opaque.[6]

Microscopically, COFs showed trabeculae or spherules of mineralized materials in a cellular fibrous connective tissue stroma. The characteristic microscopic criteria for diagnosis of COF include presence of a mixture of woven and lamellar bones and cementum-like materials in a cellular fibrous connective tissue stroma In addition, osteoblastic rimming is usually found. Variable levels of expression of fibrous and vascular components are also found. The stromal component is highly cellular to moderately cellular, prominently vascular and collagenous. Multinucleated osteoclasts-like giant cells are noted.[4]

The well-defined borders of the central COF differentiate it from the aggressive sarcomas and carcinomas. Fibrous dysplasia has a characteristic "ground glass" appearance not seen in the central COF. The radiologic differentiation of central \ COF from Gorlin cysts and Pindborg tumors is difficult; the final diagnosis is based on histologic appearance. Pindborg tumors have a high association with impacted teeth.[8]

Complete removal of the lesion at earliest possible treatment been suggested by majority of the authors.[10] Appropriate treatment for a benign fibro-osseous lesion, irrespective of its aggressive nature includes either curettage or enucleation of the lesion, until healthy margins are reached. Enucleation or curettage of the lesion is the initial treatment of choice for COF.[9] Lesions that did not produce marked deformity or obstruction at initial presentation, curettage or peripheral ostectomy alone appeared to be adequate management along with long-term clinical and radiographic follow-up.[4] Successful removal can also be achieved by local excision and enbloc resection.[9]

Excision of the tumor along with safe margins was done in the reported case. Radiotherapy is contraindicated because tumor is radioresistance and post-operative complications. Recurrence rates ranging from 30% to 58% and 0% to 28% have been described by Mintz and Velez[6] and Chang *et al*.[4] respectively. Since recurrence rate is variable therefore patients should be followed-up regularly.

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How to cite this article: Kumar A, Gupta DS, Jurel SK, Khanna R, Yadav M. Extensive ossifying fibroma of jaw. Natl J Maxillofac Surg 2014;5:224-7.

Source of Support: Nil. **Conflict of Interest:** None declared.