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CASE REPORT

Huge Ossifying Fibroma of the Maxilla

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Abstract Ossifying fibroma of bone is a central neoplasm of bone. It is more common in young adult with marked predilection for mandible. It is more common in females. Lesion is generally asymptomatic until growth produces a noticeable swelling and deformity. It presents an extremely variable roentographic appearance depending upon stage of development. This lesion is composed basically of many delicate interlacing collagen fibres, 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.

Keywords Ossifying fibroma · Maxilla · Huge

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 possible odontogenic origin, 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]. Tumour manifests as a slow-growing, asymptomatic, intraosseous mass, most frequent in females between 35 and 40 years of age [4, 5]. COFs of the mandible are common, but COFs of the maxillary sinus are rare and only a few have been reported in literature. Surgical excision is the treatment of choice [4] and recurrence is variable [6].

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Case History

A 45 year old female patient reported to our department; in August 2008 with chief complaint of a huge swelling on right side of face which was gradually increasing in size since last 4 year. Patient had a history of tobacco chewing and beedi smoking since last 30 years. On examination the swelling was 8.5 × 10.0 cm in size at right maxillary region displacing the right ala of nose, columella and septum on the left side. Superiorly the swelling displaced the lower eyelid compressing the eyeball giving a sun-set appearance of the eye. Inferiorly the swelling distorted the normal appearance of upper lip. Laterally the swelling has involved the zygomatic region and up to the preauricular region (Fig. 1). Superficial skin was normal and non-adherent to lesion but stretched with prominent vascular markings. 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 with a normal appearing mucosa but

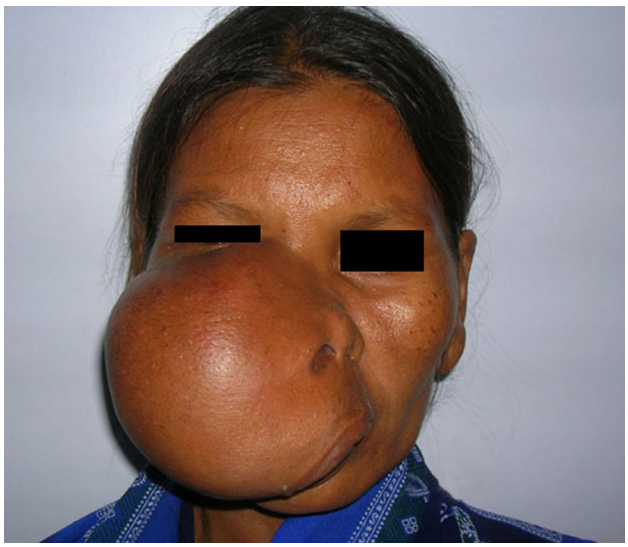

Fig. 1 Pre-operative photograph of the patient

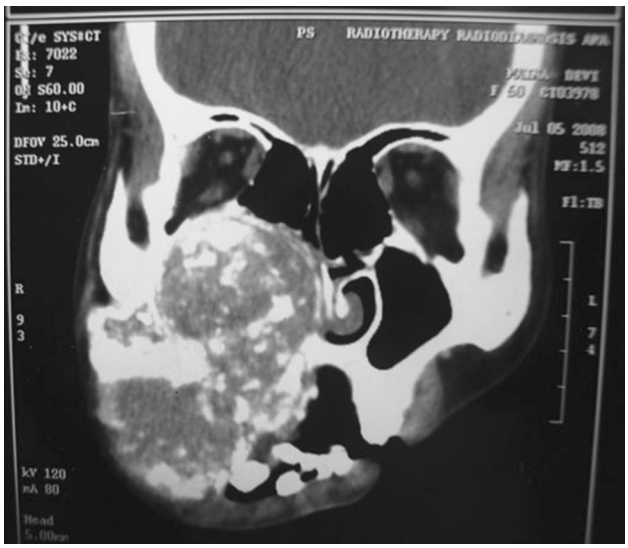

Fig. 2 Computed tomogram coronal view

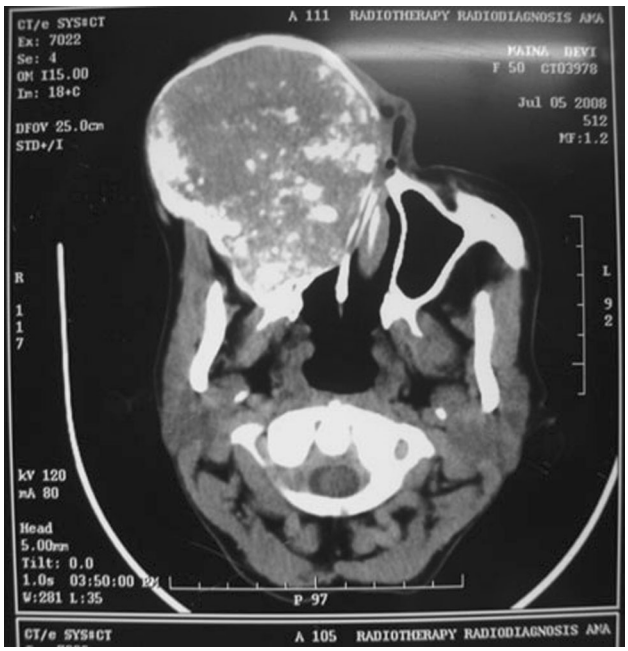

Fig. 3 Computed tomogram axial view

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. Haematological, biochemical and other investigations were within normal limits.

Incisional biopsy was done under local anaesthesia. Histological examination of specimen revealed many delicate interlacing collagen fibres, seldom arranged in discrete bundles, interspersed by large number of active proliferating fibroblasts with occasional presence of cementum like droplets. On the basis of these the diagnosis of cemento-ossifying fibroma was made.

CT scan revealed a 7.5 × 8.5 × 10 cm expansile lesion arising from 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 varying 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 intraorbital 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 was also involving anterior ethmoidal air cells, but did not cross 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 (Fig. 2, 3).

As per histological findings patients was planned for excision of lesion. After nasal intubation and under general anaesthesia we decided to go for intraoral approach. Incision was given on the most elevated point of lesion. Then the superficial mucosa was reflected using periosteal elevator until the total exposure of lesion was seen. Then the lesion was excised in toto until healthy margins of bone were seen.

Per operatively the lesion was encapsulated. It was removed with minimal bleeding and minimal injury to vital structures (Fig. 4, 5). Excised specimen was submitted for histolopathological examination, which confirms the diagnosis of ossifying fibroma (Fig. 6).

Post-operatively patient had mild post operative blackening of superficial skin on right side of face. The level of right eye was restored to normal which was elevated pre

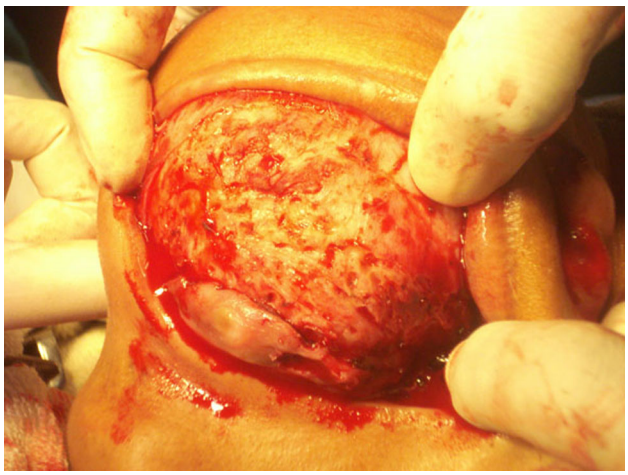

Fig. 4 Intra-operative photograph of the patient

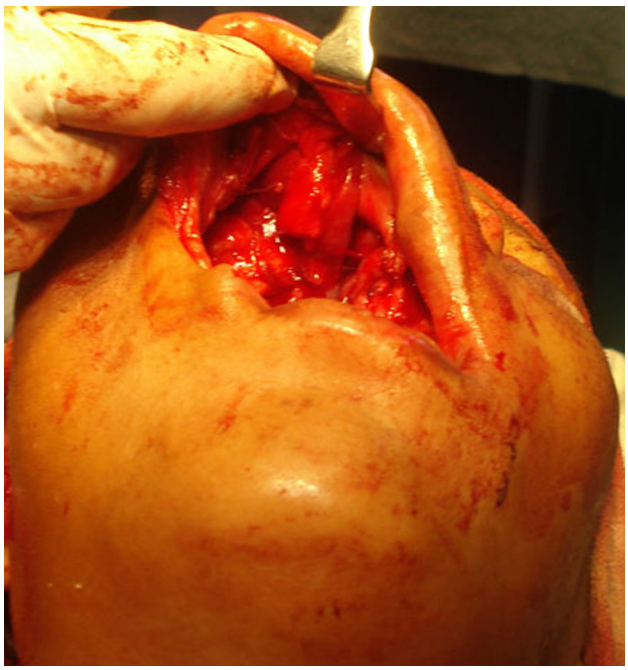

Fig. 5 Intra-operative photograph of the patient showing defect

operatively due to the massive size of lesion. Anatomic structures like alae of nose, columella, septum and upper lip were also restored to normal position (Fig. 7).

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 maxilla.

Discussion

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

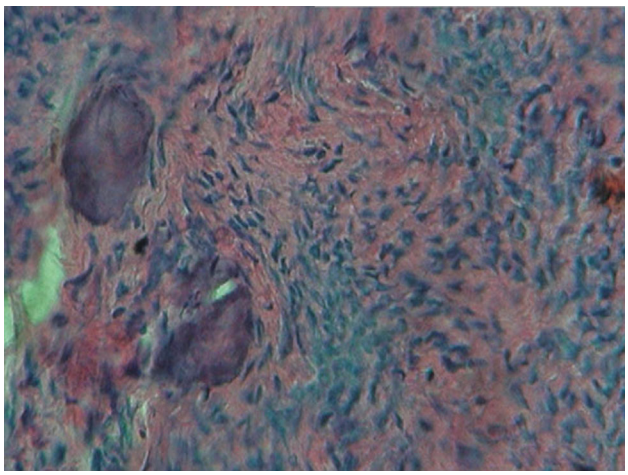

Fig. 6 Histopathology slide showing interlacing collagen fibres, interspersed by proliferating fibroblasts

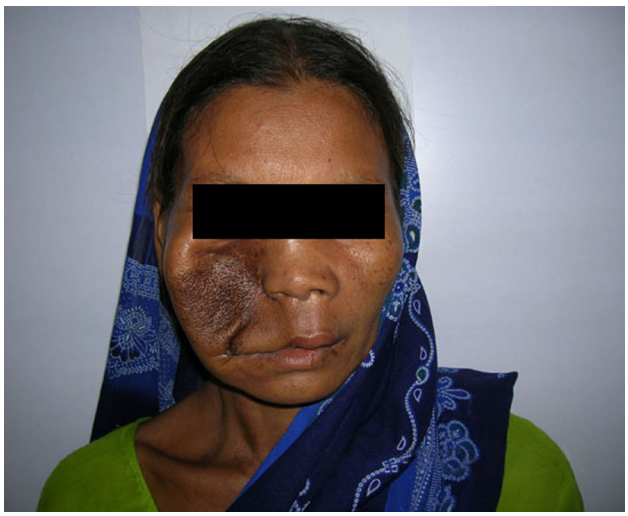

Fig. 7 Post-operative photograph of the patient

architecture by fibrous tissue, which may, mineralize in various forms like woven, lamellar 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 as central cementifying fibroma. In other cases, the calcified fragments are osteoid, these tumors are named central ossifying 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]. 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. The 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]. Root displacement has also been demonstrated in 17 and 18% of COFs reported by Eversole et al. [5] and Sciubba and Younai [9] respectively.

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 fibro-osseous 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].

Complete removal of the lesion at the earliest possible is treatment of choice, has 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. 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 tumour is radioresistance. Recurrence rates ranging from 30 to 58% and 0 to 28% have been described by Mintz et al. [6] and Chang et al. [4] respectively. Since recurrence rate is variable therefore patients should be followed up regularly.

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