Ipsilateral Maxillo-Mandibular Ossifying Fibroma

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Abstract Ossifying fibroma is a benign fibro-osseous lesion which occurs at various locations in maxillofacial region. A synchronous presentation of this lesion in both, the maxilla and the mandible is a rare occurrence. The present article reports a case of ossifying fibroma in a 45-year-old woman who manifested lesions in the right maxilla and mandible. The clinico-radiologic and histopathologic modalities of diagnosing this entity are evaluated and discussed.

Keywords Ossifying fibroma · Maxillo-mandibular · Giant maxillary ossifying fibroma · Cemento-ossifying fibroma

Introduction

To judiciously classify any lesion as a fibro-osseous lesion, its clinical, microscopic features of trabeculae or spherules of bone or cementum-like material in a cellular fibrous connective tissue stroma [5, 7, 27] and radiological features must be taken into account. The World Health Organization (WHO) in its third edition adapted the term “ossifying fibroma” (OF) instead of “cemento-ossifying fibroma” (COF) as mentioned in the earlier editions [3, 28]. OF

affecting the jaws is histologically confirmed not as an OF but only as a fibro-osseous lesion (FOL) [17]. Histopathologically, this group of lesions also includes fibrous dysplasia (FD), florid osseous dysplasia (FOD) and focal osseous dysplasia (FocOD). These lesions can be further classified as ossifying, cementifying, aggressive/juvenile form, multiple OF and familial gigantiform type [22].

OF has been reported to occur in various age groups, ranging from children to adults of old age. It is a slow growing tumor, which enlarges in size progressively to cause destruction and deformation of surrounding bone [1, 13, 19, 21]. Behaviourally these lesions are proliferative and may exhibit benign indolence to aggressive behaviour [8, 10, 11, 32]. In jaws, the mandible is affected more often than maxilla [5]. However, a synchronous occurrence of this entity in both the maxilla and the mandible is an uncommon phenomenon. We report a case of OF in a 45-year-old female who manifested lesions in both, the maxilla and the mandible on the same side.

Case Report

A 45 year old female patient was referred by a dental surgeon with the chief complaint of a painless progressively enlarging swelling on the right side of the face for the last 4 years. The patient had also noticed another progressively growing swelling on the same side albeit in the lower jaw. There was no associated pain, paraesthesia, or tenderness. Extra oral examination revealed a well-defined massive spherical swelling in right mid-facial region extending and involving the lower part of face. Superiorly the extent of the swelling was from the right infraorbital rim to the right commissural area. Medio-laterally, the lesion extended from bridge of the nose up to the

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Fig. 1 Patient with giant maxillary ossifying fibroma (*frontal view*)

ala region, involving the zygomatic region. The size of the swelling was approximately 4×5 cm. Additional findings included an apparent drooping of the upper lip with a widened philtrum, distorted appearance of nose and external nares along with premature closure of upper eye. Skin overlying the swelling was normal in appearance without any surface changes. The swelling was firm in consistency, fixed to the underlying mucosa with no associated temperature change or tenderness (Figs. 1, 2).

Intra-orally, the maxillary lesion revealed obliteration of the buccal and labial vestibule on the right side with the absence of all maxillary posterior teeth on the right side. Fine capillaries were seen on the overlying mucosa resulting from an underlying pressure exerted by this space

Fig. 2 Patient with profile view

occupying lesion. There was an expansion of the buccal and palatal cortical plates. Moreover, the remaining maxillary and mandibular anterior teeth were also mobile. Further enquiry revealed a gradual loss of molars over a period of time with progressive enlargement of the lesion. Teeth indentations over the swelling were noted caused by the impingement of mandibular teeth.

Another swelling of approximately 3×4 cm was seen in the right mandibular premolar region, with missing 2nd premolar and molars. No associated pain, tenderness or paraesthesia was noted on palpation. Mouth opening of the patient was adequate with a patent nasal airway (Fig. 3).

A complete hemogram, serum chemistry including serum calcium, phosphorus and alkaline phosphatase was advised, which was within normal limits. An orthopantomograph and a 37° occipitomeatal view showed complete opacification of right maxillary sinus with generalized inhomogeneous increased thickness of sinus floor. Axial CT through the maxilla was prescribed for the extent and internal details of the lesion. CT showed presence of two distinct well corticated space occupying lesions in maxilla and mandible on the right side.

The maxillary lesion was $8.16 \times 5.38 \times 5.76$ cm in the antero-posterior, transverse and supero-inferior dimensions respectively. It was a large mixed radiolucent-radio-opaque lesion, walled in an irregularly thick sclerotic border, located at the alveolar process of both upper and lower jaws. The superior margin reached the orbital floor with no involvement of intraorbital structures. The lesion extended antero-posteriorly from anterior nasal aperture up to the level of lateral pterygoid plate. Medio-laterally, the lesion extended from the lateral part of the nasal cavity up to the maxillary buttress covering the maxillary sinus. The right maxillary sinus and the right nasal cavity were completely obliterated due to ingress of lesion and subsequent mucosal thickening in conchae region. The lesion was well defined

Fig. 3 Patient with maxillary and mandibular lesions

with distinct cortical outline. Scalloping was observed in the area where presence of other bony structures caused an impediment in growth of this expansile lesion viz. zygomatic region. The internal structure comprised of foci of calcification, more abundant in the area of the obliterated maxillary sinus (Fig. 4).

A second distinct mixed radiolucent–radio opaque lesion was noted in the right mandibular region distal to 1st premolar (Fig. 5). The lesion was expansile in nature with a nidus of calcification observed at the level of alveolar crest suggestive of the fact that its origin was from a tooth bearing area. Moreover, a comparable expansion of buccal and lingual cortical plate in both the directions was evident. The lesion measured $1.93 \times 1.82 \times 1.37$ cm in the antero-posterior, transverse and superio-inferior dimensions respectively (Fig. 6).

A provisional diagnosis of OF was made based on the clinical findings and findings on CT. Differential diagnoses include florid cemento-osseous dysplasia, ossifying and cementifying fibroma, osteoma arising from sinus, with concurrent presence of cementoblastoma. Biopsy was performed on both lesions; histopathological examination confirmed the diagnosis of COF. Pathological examination revealed a composite tissue consisted of lamellar bone trabeculae, scattered throughout a fibrous stroma. Inside the fibromatous stroma, fibroblast like and osteoclastic cells were present. On haematoxylin–eosin staining few areas of plump fibroblast and presence of mature bone with osteoblastic rimming was observed. Irregular eosinophilic

Fig. 4 CT scan (axial section) showing extent of maxillary lesion

Fig. 5 CT scan (axial section) showing mandibular and maxillary lesions

Fig. 6 Hypercellular stroma with ossified tissue

globules of calcified material closely resembling acellular cementum like tissue were also seen confirming the diagnosis of OF (Fig. 7).

Patient was treated by excision and enucleation of the tumor mass, after exposing the tumor through standard Weber Ferguson approach (Fig. 8). Since the maxillary tumor was large in size it was not possible to remove the tumors enmass without jeopardizing the adjacent vital

Fig. 7 Ossified tissue in higher magnification

Fig. 8 Exposure of tumor through Weber Ferguson approach

structures, so the tumor was excised and enucleated completely as it is easily separable from adjacent tissue (Fig. 9). Mandibular tumor was excised completely in toto. For maxillary defect an obturator was made. Although patient was advised to have regular follow up, but patient did not reported after first 3 months of follow up (Fig. 10).

Discussion

Waldron [28] classified FOLs broadly into three categories—FD, reactive lesions arising in the tooth bearing area from periodontal ligament and fibro-osseous neoplasm. Montgomery [15] was the first to report a case of OF of jaws in 1972. Since then, cementifying fibroma and COF have been reported as histologic variants. Eversole [5] further classified COF into five types namely: ossifying form, cementifying psammomatoid form, aggressive (juvenile) form, multiple OF and familial gigantiform cementoma. The term “COF” is used to

Fig. 9 Excised maxillary and mandibular tumor

Fig. 10 Post operatively after 2 weeks

describe fibrous lesions containing calcifications with a strong similarity between the bone and cementum. The WHO in its third edition, adapted the term “ossifying fibroma” (OF) instead of the previously mentioned “cement-ossifying fibroma” [3].

Although WHO and some authors regard the CF as an odontogenic tumor and consider OF separately as non-

odontogenic neoplasm, there is general agreement that CF and OF represent only histologic variants of the same lesion (Stergiou [23]). OF is limited to the bones of craniofacial complex although extragnathic OF has been described in long bones with a similar histology. A number of authors have suggested that ossifying and/or cementing fibromas are benign FOLs originating in the periodontal ligament—as a result of which they would constitute histological variants of the same pathological process [4, 6, 9, 25, 30, 31].

OF is an uncommon, benign, monostotic, well-defined, unilocular or multilocular fibrous-osseous tumor, arising from the periodontal ligament [2, 25]. It most frequently occurs in female patients (age range 10–59 years, mean 32 years) with an incidence peak in the third and fourth decades [25]. However, the average age varies according to the source of origin of the lesion. Prime incidence is in the second and third decade, whereas others reports third and fourth decade as a principal period of incidence. 70 % of cases of OF involve the mandible but a significant percentage (22 %) was found in the molar region of the maxilla, ethmoidal and orbital regions and exceptionally in petrous bone [20, 24].

A noticeable deformity arises when the lesion involves the jaws or the orbit because of the expanding nature of disease. These lesions however, go unnoticed for many years when they arise in bones that underlie the sino-nasal lining. Growth rate is often slow and the lesions progressively enlarge typically up to 1–2 cm in size, occasionally growing at an alarming rate to reach more than 4 cm causing jaw expansion [16]. A juvenile variant can show aggressive growth [12]. These rapidly growing forms of OF have been termed as juvenile, but as not all patients are young, the term aggressive OF is preferred [29].

The characteristic radiographic features of OF are expansion and margination, demarcation or cortication. They maintain a spherical shape, expand the surrounding cortical bone without cortical perforation and may cause tooth divergence or even resorption with gradual loss of teeth. These findings were also seen in the present case. Early lesions are radiolucent, as the lesion matures; confluent floccular opacities become evident and are delineated from the adjacent lesion by a radiolucent halo. Tumors arising in the maxillary, ethmoidal or sphenoidal sinus usually exhibit a round or oval outline and expand into the sinus cavities. In the present case a complete obliteration of the maxillary sinus was observed with these confluent floccular opacities. The mandibular lesion also depicted a similar pattern wherein the epicenter was located at the alveolus.

A typical OF shows fibroblastic cellularity with bony trabeculae (woven or lamellar) rimmed by osteoblasts [12]. Many OF are predominantly or exclusively composed of small ovoid calcifications referred to as cementicles,

psammoma bodies or ossicles. Hypocellular myxoid regions alternating with clonal zones of active osteogenesis composed of thin rudimentary osteoid trabeculae, rimmed by a large polygonal and angulated osteoblasts may be seen. In other regions there may be small confined areas with multinucleated giant cells. In the present case irregularly shaped calcification (cementicles, psammoma and ossicles) with zones of hypo cellular myxoid regions in a hypercellular fibrous connective tissue stroma was seen.

Clinical and radiological differential diagnoses of the maxillary lesion include lesions that contain radio-opacities within a well-defined radiolucent mass. They include focal cemento-osseous dysplasia, focal sclerosing osteomyelitis, florid cemento-osseous dysplasia, osteoblastoma, chondrosarcoma or osteosarcoma, giant cell lesions, calcifying epithelial odontogenic tumor/Pindborg tumor [15]. Focal cemento-osseous dysplasia is not an expansile lesion and does not have a progressive growth potential, while osteoblastoma is more frequently discovered in the inferior aspect of the mandible. Aggressive osteoid osteoma, sarcoma and carcinomas can be differentiated on their growth potential and borders as these do not exhibit well-defined outline. Giant cell granuloma may also resemble an OF clinically, with both types commonly occurring anterior to the molars and presenting as pedunculated or sessile masses. However, the peripheral giant cell granuloma can be ruled out on the basis of the size of the lesion (0.5–1.5 cm), its pedunculated nature, and the presence of peripheral cuffing and giant cells on histopathological examination. Peripheral cuffing, due to superficial erosion of the bone, is the distinct radiographic feature. In calcifying epithelial odontogenic tumors, the common site of occurrence is the molar area, whereas the OF is generally seen anterior to the molars. Calcifying epithelial odontogenic tumor is also generally associated with an unerupted or an impacted tooth. Initial radiographic features of both the lesions are common (both being radiolucent) but as the lesion progresses it takes on a ‘honeycomb’ or ‘driven snow’ appearance. Histologically it can be differentiated by the presence of polyhedral epithelial cells as well as multinucleated giant cells. Radiology is central to their diagnosis because the pathology for all fibro osseous lesions is similar; although they range widely in behavior [30]. FD has a characteristic “ground glass” appearance not seen in the central OF. The radiologic differentiation of central OF from Gorlin cysts and Pindborg tumors is difficult; the final diagnosis being based solely on histologic appearance [12, 14].

Although incisional biopsy is diagnostic of OF, it does not provide any clue as to its aggressiveness. Therefore, any large maxillary lesion should not be assumed to be aggressive, but it is prudent to treat such lesions by thorough enucleation followed by careful curettage of tumor cavity walls—as was also done in the present case. The patient should be observed very carefully to verify the

complete resolution since recurrence is the most distinguishing feature of an aggressive OF. Recurrent lesions of belligerent variety require a more aggressive approach. In the present case, the entire tumor was removed including involved regions of the orbital floor and maxillary sinus walls. OFs are usually excised easily at surgery, but a maxillary central OF is more difficult to remove completely than a mandibular central OF [12, 18, 26]. A complete excision of the tissue is the method of choice; however, recurrence is the only feature that is predictive of the effective management.

Conclusion

Although OF is a rare entity by itself, its presence in multiple locations in a patient is extremely uncommon, and these different lesions may appear at different point of time in a patient life time, so patient needs to be observed after treatment to look for any recurrences as well as for any new lesion over a prolonged period of time. Whether maxillary lesions grow at a more rapid rate in a small period of time than mandibular lesions is not known. However it is necessary to treat them at the earliest possible, so that surrounding vital structures can be preserved. Also a continued clinical and radiological monitoring is required before deciding on osseous reconstruction with sufficient guarantees of non-recurrence.

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