Central ossifying fibroma of the Mandible – Report of a rare case

Abstract
Central ossifying fibroma is a relatively rare, benign, non-odontogenic tumor of the jaw, a subdivision of fibro-osseous lesions. The lesion originates from periodontal membrane and is usually seen in tooth bearing areas particularly of the mandible. This bone tumor consists of highly cellular, fibrous tissue that contains varying amounts of calcified tissue resembling bone, cementum or both. Correct differentiation of this lesion from other fibro-osseous lesions is vital as their management varies from none to surgical enucleation or complete resection. Present report describes the comprehensive surgical management of a large ossifying fibroma of the mandible in a young patient with segmental resection & reconstruction using a stainless-steel reconstruction plate.

Key words: Ossifying fibroma, tumor, fibro-osseous lesion, resection
Introduction
There is a group of pathological entities occurring in the bones of the jaws and skull, which in current medical terminology are commonly grouped together under the generic term fibro-osteocemental lesions[1]. They represent a diverse group of diseases characterized by the replacement of normal bone with a variably collagenous connective tissue matrix containing trabeculae of new bone and, in some lesions, cementum like material. Ossifying fibroma is designated as a benign form of fibro-osseous lesion with well circumscribed, slow growing and sharply defined margins and a radiolucent peripheral component. It shares identical clinical, radiographic and histopathological features with a lesion previously termed cementifying fibroma[2]. Both these lesions are considered one and the same nowadays. In view of microscopic similarities with fibrous dysplasia and the cemento-osseous dysplasias, some investigators regard this lesion as an example of a localized dysplastic process in which bone metabolism has been altered. We present a case of large ossifying fibroma of mandible in a young patient comprehensively managed with segmental resection & reconstruction.

Case Report
A 20 year old male patient reported to the maxillofacial out patient department with the chief complaint of gradually increasing swelling over left side of lower jaw since 2 years. Swelling was insidious in onset without any history of trauma, dental pain or treatment. There were no associated symptoms of pain, pus discharge or paresthesia. Aspiration of the swelling was attempted but was unsuccessful.

Figure-1: Pre-operative intraoral view showing left mandibular lesion with expansion of buccal and lingual cortices

His past medical history, family and personal history was non-contributory. Extraoral examination revealed diffuse swelling in left parasymphysis-body region of mandible, extending from angle of mouth till inferior border. It was well defined, bony hard and non tender. Submandibular lymph nodes were palpable, enlarged and tender. Intraorally, a 3.5 cm x 2.5 cm swelling was seen in left mandible extending from canine to first molar region and obliterating the buccal vestibule. Swelling was bony hard and non-tender with expanded buccal and lingual cortices (Figure-1). No associated discharge or tooth mobility was present. Radiographic examination in the form of OPG revealed a well defined, multilocular, radiolucent lesion extending from right mandibular central incisor to left first molar region. Flaring of roots of premolars was noted (Figure-2A). CT scan showed a well-defined, expansile, multiseptated lytic lesion
approx. 3.6×2.8 cm in left mandibular body region with bicortical thinning without perforation (Figure-2B).

*Figure-2A: Pre-operative OPG showing multilocular radiolucency with flaring of premolar roots*

*Figure-2B: Axial & coronal CT views showing expansile, multiseptated, lytic lesion in left mandible*

Incisional biopsy of the lesion was performed under local anesthesia which was suggestive of fibro-osseous lesion. Segmental resection of the left half of mandible was planned intraorally under general anesthesia along with primary reconstruction with a stainless steel reconstruction plate.

*Figure-3: Surgical exposure of the lesion*
A submarginal incision was made extending from the region of mandibular left central incisor to second molar with mesial and distal releasing incisions. Mucoperiosteal flap was reflected to expose the lesion which was well circumscribed with expansile buccal cortex and irregular surface (Figure-3). Mandibular left central incisor and second molar were extracted and vertical osteotomy cuts made through the respective sockets. Muscle attachments on the lingual surface of mandible were stripped and segmental resection completed. A stainless steel reconstruction plate was adapted to match the contour of the resected mandible. Temporary intermaxillary fixation was done and the plate was fixed using 2.5 mm diameter, 10 mm long stainless steel screws (Figure-4A and B). Intermaxillary fixation was later released and wound closed in single layer using 3-0 vicryl sutures.

**Figure-4A: Reconstruction plate fixed after segmental resection**

![Reconstruction plate fixed after segmental resection](image)

**Figure-4B: Post-operative OPG showing well adapted reconstruction plate**

![Post-operative OPG showing well adapted reconstruction plate](image)

Post operative course of the patient was uneventful and he was discharged on 12th post operative day. The histopathology report of the specimen confirmed the diagnosis of ossifying fibroma.

**Discussion**

Ossifying fibroma is considered by most to represent a benign neoplasm arising from undifferentiated cells of the periodontal ligament tissues. Due to its origin from multipotent mesenchymal cells, it can produce bone, cementum, spheroidal calcifications and fibrous tissues. In 1971,
WHO classified four types of cementum containing lesions: fibrous dysplasia, cementifying fibroma, ossifying fibroma and cemento-ossifying fibroma[1]. According to the second WHO classification, benign fibro-osseous lesions in the oral and maxillofacial region were divided into two categories, osteogenic neoplasm and non neoplastic bone lesions; cementifying ossifying fibroma belonged to the former category [2]. However, the term “cementifying ossifying fibroma” was reduced to ossifying fibroma in the new WHO classification in 2005[3]. Ossifying fibroma is typically a slow-growing, well circumscribed, expansile lesion that replaces normal bone as it enlarges. Most lesions are asymptomatic when detected. These lesions commonly appear between 20-40 years of age, with females five times more frequently affected than males [4]. Mandible is most commonly involved in most of the cases, with rare exceptions, lesions arise in the tooth-bearing regions of the jaws, mainly the mandibular premolar or molar region. The lesions are usually firm, non-tender on palpation. In maxilla, they may cause cortical expansion, obliterating the buccal sulcus and maxillary sinus. Extension into the nasal cavity and orbital floor may lead to epistaxis and epiphora. Expansion of both cortical plates may be seen in case of mandibular lesions[5]. Extension of tumor mass into the ramus of mandible and involvement of the inferior border may cause paresthesia of inferior alveolar nerve. Patients with clinically progressive lesions may seek treatment due to pain, paresthesia and facial asymmetry. When the lesion arises in children, it has been named the juvenile aggressive cemento-ossifying fibroma, which presents at an earlier age and is more aggressive clinically and more vascular at pathological examination[6]. Radiographically, two major patterns of ossifying fibroma are seen[7]:

**Expansile unilocular radiolucency with or without opacifications**

The radiographic appearance is variable, depending on the degree of maturation and the amount of mineralization present. Early lesions are typically present as unilocular or multilocular radiolucencies. This initial radiolucent stage gradually progresses to a mixed radiolucent-radiopaque stage as matrix material is deposited and mineralized in the lesion. Fully mature, long-standing lesions appear as dense, radiopaque masses surrounded by a thin, well-defined, regular, radiolucent rim. As lesions enlarge, they may displace adjacent teeth and, less commonly, cause root resorption. Histopathologically, the tumor consists of a collagenous stroma containing variable numbers of uniform spindled or stellate cells. The degree of vascularity is variable. One feature that distinguishes ossifying fibroma from fibrous dysplasia is the focal presence of osteoblasts along the surface of the bone deposits. Basophilic, spherical calcifications and anastomosing trabeculae of cementum-like material are also frequently present. The observation of this type of calcified matrix has led most investigators to conclude that ossifying fibromas and cementifying fibromas are the same lesion.

The well-circumscribed nature of ossifying fibroma is evident at the time of biopsy or surgery, when the lesion may be easily removed from the surrounding bone in one piece or several large fragments. An intraoral approach for the surgical excision of the tumor by enucleation is the preferred method of management. Adjacent normal structures including teeth, neurovascular elements and bone should be preserved whenever possible. When large lesions are excised and the potential risk for postoperative fracture is present, intermaxillary fixation should be considered during the initial healing stages. Recurrence is described rarely following complete surgical excision. Recurrent lesions can be managed conservatively unless they are large causing extensive destruction of bone. These more
extensive lesions, such as the one presented, whether recurrent or primary, may require surgical resection and bone grafting as definitive treatment.

References