INTRODUCTION

Cemento-ossifying fibroma (COF) is the most common benign fibro-osseous neoplasm of the jaws, which can be either unilocular or multilocular. This benign, locally destructive tumour tends to occur in the maxillofacial region and can cause marked cosmetic and functional deformity after attaining large size especially if left untreated.¹ The central cemento-ossifying fibromas of the mandible are common, however, unusual in the maxilla.²,³ It should be distinguished from fibrous dysplasia of bone and certain other fibro-osseous lesions that do not represent true neoplasm. A close histogenetic relationship exists between the central cemento-ossifying fibroma and the central ossifying fibroma. However, these two lesions represent the same basic neoplastic process.⁴ The only difference found between these two entities is the formative cell involved and its final product being cementum in one case and bone in the other.

This tumour generally occurs in young and middle-aged adults. There is a marked predilection for the female gender and the female: male ratio is 5:1. The mandible is more commonly involved than the maxilla and mandibular premolar-molar area is the most common site. In the early stages, the cemento-ossifying fibroma appears as a radiolucent lesion with no evidence of internal radio-opacities.² As the tumour matures, there is increasing calcification so that, the radiolucent areas become flecked with opacities until the lesion appears as a mixed radiopaque mass.³,⁴ Displacement of adjacent teeth is a common finding. Another additional diagnostic feature is that, there is a centrifugal growth pattern rather than a linear one, therefore, the pathology grows by expansion equally in all directions and presents as a round tumour mass. Its circumscribed nature, permits enucleation as treatment option especially, when of small size. This patient, clinically presented with a huge, irregular with a few small and large growths all presenting in a single mass, clinically appearing as a mandibular dumbbell tumour. However, larger lesions which destroy the surrounding normal bone often require surgical resection and bone grafting, which was planned for this case.

CASE REPORT

A 32 years old male patient reported at the Department of Oral and Maxillofacial Surgery, Civil Hospital, Karachi/ Dow Medical College with complaint of painless and gradually increasing disfigurement of his lower face for the last 3 years. He was apparently asymptomatic 4 years back, when he developed a small swelling in the right posterior mandibular region. This had gradually increased in size. He was surgically operated in the past for this problem, but was not having any supportive records.

The extra oral clinical examination showed a hard, smooth-surfaced bony mass present with irregular and large multi-nodular growths, with some soft areas on the
right side of the face. The intra oral clinical examination showed a tumour having smooth oral mucosal surface with hard bony areas. This had caused buccal and lingual cortical expansion of the mandible, starting from chin upto the retromolar area. Clinically, this tumour appeared like a dumbbell mass. All his posterior teeth were absent on the same side. However, few teeth were also present on his upper and lower jaws. His past social, medical and dental history was insignificant. The orthopantomograph (OPG) showed a very large, mixed radio-opaque and radiolucent lesion (Figure 1). Clinical and radiological differential diagnosis included ameloblastoma, myxoma, and cemento-ossifying fibroma.

An intraoral Incisional biopsy was done with informed consent, under local anaesthesia. C.T scan was done with contrast medium, showing bone destruction. The tumour extended in the right infra temporal and para-pharyngeal space, causing compression of the lateral para-pharyngeal wall, tongue and floor of the mouth. This tumour measured, 9.0 x 7.5 cm and extended 8.5 cm in cranio-caudal dimension. Biopsy report suggested cemento-ossifying fibroma of mandible. Elective surgery was planned under general anaesthesia for en bloc tumour resection and reconstruction. Therefore, a right side submandibular incision from chin upto the mastoid process was given, avoiding a split lower lip. This incision started from, the labiomental groove, below his lower lip involving the chin and extended upto the right mastoid process. Mandibular en bloc tumour resection was done from the right midline and the entire tumour mass involving the condyle-coronoid processes was removed (Figure 2). The tumour weighed approximately 300 grams. Intraoral and extra oral seals were made by closing the soft tissues in layers.

Drain tube was not used in the surgical bed, instead it was packed with apx. 30 inches long Bismuth Iodoform Paraffin Gauze Pack (BIPP). Routine intravenous medication was used. The BIPP pack was removed in small increments of 06 inches on 3rd, 5th, 7th and 10th postoperative days. The nasogastic tube for feeding was also not used; instead disposable plastic oral straws were advised. Reconstructive surgery was planned after 3 months. Post operative recovery phase was uneventful (Figure 3). He was discharged on the 12th post-operative day and came back after 3 weeks for follow-up with great improvement in his facial deformity and a cheerful face. So far, he has not reported back.

**DISCUSSION**

Fibro-osseous lesions are a heterogeneous group of benign lesions of unknown aetiology affecting the jaws and other craniofacial bones. COFs, fibrous dysplasias, ossifying fibromas, and cementifying fibromas were defined by MacDonald in 2004. COF is the most common fibro-osseous lesion encountered by an Oral Pathologists and perhaps, it has more synonymous than any other jaw lesion. Unlike fibrous dysplasia, COF is considered as an osteogenic neoplasm manifesting as a slow growing, asymptomatic, well-defined unilocular or multilocular intraosseous mass.

Central cemento-ossifying fibromas are asymptomatic well-defined, solitary radiolucencies with scattered radiopaque foci that maintain a spherical shape, expand
the surrounding cortical bone without cortical perforation, and may cause tooth divergence.\(^5\) Similar behaviour was also observed in this case, associated with massive medio-lateral cortical tumour expansion with bone destruction.

COF may also manifest as Stafne’s bone cyst, central giant cell granuloma, residual cyst or odontogenic keratocyst. Delayed presentation may be due to many reasons. These lesions are relatively uncommon and unlikely to be seen and treated by the general dental practitioner and unrelated medical specialist. The understanding of this oral pathology to diagnose and treat when encountered was one of the reason for late presentation, further hampered by the financial aspects of investigations and treatment.

The authors’ interpretation for this case was subjective and based on visual comparison of the available OPG X-ray. Since the actual oral pathology of this patient was not entirely surgically removed earlier. The description of histological characters may sometimes be difficult to see on incisional biopsy alone, especially if proper representative tissue specimen is not taken. CT scan is necessary to check the anatomical extent of the lesion for elective surgical procedure. Zupi et al. reported two features that may help in distinguishing aggressive cemento-ossifying fibroma (ACOF) from central COF.\(^6\) First clinically, the ACOF occurs at a far lower mean age than the central COF. Secondly, the histological pattern of the ACOF seems to be unique in being highly cellular with entrapped osteoblasts. The characteristic feature of aggressive type of COF are its early occurrence, high incidence in children and young adults. It is more common in maxilla, aggressive behaviour which causes marked destruction of surrounded tissues, which become symptomatic in a very short course of its evolution, causing pain, paresthesia, bone perforations and high tendency to recur after removal. However, considering the age, radiographic appearance and the growth behaviour of this case, it fits under the COF category, being the non-aggressive type. Review of the local Pakistani maxillofacial literature found no COF case of such massive size and technique, being operated successfully by saving the splitting of his lower lip reported, although International Journals document advantages of this incision.\(^7\)\(^8\) Lower lip-split incision produces peri-oral scars, while the modified submandibular incision in the neck crease, without splitting of lower lip gives better aesthetics and avoids lip scar. Saving the lower lip from splitting also saves blood loss and stitching time during surgery.\(^9\) Bismuth iodoform paraffin paste (BIPP) has been used in dressings in ENT, dental, and neurosurgical practices. BIPP dressing used can be left as a specialized, cost effective and easy to use dressing to keep the area infection free. BIPP also has the potential advantage to stop most of the hemmarrages.\(^10\) The resected mandibular surgical bed in this case was packed with BIPP so, that it can be left for long periods to prevent bleeding, infection and hematoma formation. Regarding this case, long standing tumour history demonstrated a slow growing, aggressive tumour, yet with no signs of vital structures involvement. Considering the age of the patient, this case posed a dilemma as to whether to enucleate it or to perform an extensive-aggressive and radical surgery, so we opted for \textit{en bloc} resection due to its huge size, cosmetic deformity and clinical presentation.

**REFERENCES**


