INTRODUCTION

The fibro-osseous lesions of the maxillofacial bones represent a diverse group of pathologic conditions. Though, there is a substantial overlap of the histopathologic findings in the subclassification of these lesions, their management varies depending upon the actual disease process. Regardless of the type, they demonstrate replacement of normal bone by the fibrous connective tissue with admixture of mineralized product, including osteoid, mature bone and/or cementum-like calcifications. This group includes developmental, reactive or dysplastic lesions as well as neoplasms [Table 1].[1]

Clinically, they may vary from cosmetic or functional disturbances to completely asymptomatic lesions recognized on routine radiographs.

Radiographically, they may vary from solitary to multifocal or multiquadrant involvement; ill-defined to well-demarcated; completely radiolucent to mixed radiolucent radio-opaque to radio-opaque or ground glass appearance; monostotic (involving one bone) to polyostotic (involving more than one bone) and may or may not be associated with tooth-bearing region (or root apices). Grossly, the lesions may vary from smooth to gritty or may have a sandpaper-like appearance. Diagnosis of a specific lesion should only be made on the basis of full consideration of clinical, radiological and histopathological features.

Multiquadrant involvement with mixed radiolucent-radio-opaque lesions is a common finding in florid osseous dysplasias, which have a preponderance of occurring in black females. The prevalence of such lesions in Asian males is rare and only one case of florid cemento-osseous dysplasia has been reported in Indian male till date.[2]

CASE REPORT

A 26-year-old Indian male reported with mild discomfort and tenderness associated with a swelling in mandibular left posterior region since two to three months in molar region. The patient presented with bad oral hygiene with multiple decayed teeth and gave a history of multiple tooth extractions in the maxillary and mandibular regions. The overlying mucosa was normal without any signs of inflammation. Palpation revealed unilateral buccal expansion on the posterior left mandible.
Orthopantomogram showed a lobular radio-opaque mass associated with apex of impacted left third molar near the angle of the mandible. The inferior alveolar canal was pushed down. Also, there was a radio-opaque mass with a halo rim in the body of mandible on the same side. On the right side, multiple asymptomatic radio-opaque masses with radiolucent halo were evident in molar region [Figure 1]. The mass on the right side did not produce any symptoms and was found only on radiographic examination.

Table 1: Classification of benign fibro-osseous lesions of the craniofacial complex

| Bone dysplasia                        | Fibrous dysplasia                  |
|---------------------------------------|------------------------------------|
|                                       | Monostotic                         |
|                                       | Polyostotic                         |
|                                       | Polyostotic with endocrinopathy     |
|                                       | (McCune-Albright)                   |
|                                       | Osteofibrous dysplasias             |
|                                       | Osteitis deformans                  |
|                                       | Pagetoid heritable bone dysplasias of childhood |
|                                       | Segmental odontomaxillary dysplasia |
| Cemento-osseous dysplasias            | Focal cemento-osseous dysplasia     |
|                                       | Florid cemento-osseous dysplasia    |
| Inflammatory/reactive processes       | Focal sclerosing osteomyelitis      |
|                                       | Diffuse sclerosing osteomyelitis    |
|                                       | Proliferative periostitis          |
| Metabolic disease: Hyperparathyroidism | Ossifying fibroma NOS               |
| Neoplastic lesions (ossifying fibromas)| Hyperparathyroidism jaw lesion syndrome |
|                                       | Juvenile ossifying fibroma          |
|                                       | Trabecular type                     |
|                                       | Psammomatoid type                   |
|                                       | Gigantiform cementomas              |
| **NOS:** Not otherwise specified      |                                    |

Incisional biopsy was performed for the diagnostic purpose. Grossly, the tissue was creamish yellow to brown, gritty and firm to hard.

Histopathologically, the outer cortex showed thick linear trabeculae of woven bone and fibrocellular connective tissue stroma. The bone exhibited lacunae containing osteocytes and irregular basophilic reversal lines. The connective tissue consisted of collagen fibers, plump fibroblasts and few blood vessels.

The inner cortex revealed dense mass of woven bone with irregular basophilic reversal lines and scant fibrocellular stroma. In few areas, lacunae were empty. Few dysmorphic cementum like masses (cementoid) with brush border were also seen within the stroma. The connective tissue consisted of bundles of collagen fibers, fibroblasts and few blood vessels. Areas of multinucleated giant cells adjacent to bony and cementoid area were also evident [Figures 2-4]. Areas of hemorrhage were also noted [Figure 5].

Various lab investigations like complete blood count, random blood glucose levels, serum calcium, serum phosphate and serum parathyroid were carried out and were shown to be within the normal limits. Serum total calcium level, serum phosphate level and serum parathyroid level was 10 mg/dl, 3.2 mg/dl and was 25 pg/ml, respectively.

Thus, to summarize, bilateral mandibular radio-opacities surrounded by a radiolucent rim with evident masses of woven bone and dysmorphic cementum like calcifications were seen. Clinical swelling was evident only in the left posterior quadrant. After correlating the clinical, radiographic and histopathological features the lesion was diagnosed as florid cemento-osseous dysplasia.

Patient was advised for regular follow-ups as these lesions are asymptomatic and does not require resection.
DISCUSSION

A swelling with mild discomfort and tenderness of short duration in the posterior mandible of a 26-year-old male may vary from an infected, inflammatory lesion to the presence of a malignancy invading the surrounding tissues. Rarely does it occur as a result of a benign quiescent lesion, which may need not undergo extensive surgical treatment. The clinical differential diagnoses in the present case included infected dental cysts, chronic osteomyelitis, cementoblastoma, osteoblastoma and jaw malignancies due to nerve impingement as osteosarcoma and intraosseous squamous cell carcinoma. Radiographically, our case presented with mixed radio-opaque radiolucent lesion and hence, differential diagnosis included complex odontome, cemento-ossifying fibroma, fibrous dysplasia, cementoblastoma, osteoblastoma and cemento-osseous dysplasia.

Infected dental cysts clinically present as tender swellings of the jaws, hence was also considered. Radiographically, the lesion presented as a radio-opaque mass and since cysts appear as radiolucent lesions in jaws with distinct borders this differential was excluded.\[3,4\]

Condensing osteomyelitis can occur at any age though uncommon after third decade and produces swelling. It is more prevalent in mandible than maxilla. But, cases of chronic osteomyelitis are associated with long standing infected tooth. The patient had a history of carious extracted molars in left mandible which lead to consideration of condensing osteitis as a probable diagnosis. But, it is a focal lesion with involvement of the bone surrounding and extending below the apex of the teeth. In contrast our case showed multiple lesions with bilateral jaw involvement without necessary relation to the tooth.\[3,4\]

Complex odontome presents radiographically as irregular calcified material surrounded by a narrow radiolucent rim and may be associated with pain when secondary infection occurs, but histopathologically shows the presence of enamel, dentin, pulp tissue and cementum.\[5\]

Cemento-ossifying fibroma presents radiographically as a solitary spherical or oval shaped mixed radiolucent radio-opaque lesion with well-delineated margins and centrifugal growth pattern, which at a later stage may attain larger size.\[6\]

Fibrous dysplasia presents radiographically as fusiform non-demarcated radio-opacity giving typical ground glass appearance. The indistinct margins gradually blend into normal bone but on histopathological examination evenly spaced irregular shaped trabeculae with characteristic Chinese letter appearance, stellate osteoblasts with osteoblastic rimming and trabeculae having wide osteoid seams are evident.\[6\]

A characteristic histopathological finding of Paget disease is a pattern of irregular basophilic reversal lines mimicking a mosaic; due to repeated destruction and repair.\[6\] Despite this close histopathologic resemblance in the present case, Paget’s disease has been excluded as a diagnosis, as the clinical and radiographic features are contraindicating the same. Paget’s
The use of the term dysplasia refers to abnormal processes that continue, the calcified tissues grow larger and fuse to form a dense sclerotic mass. Both cemental and osseous terms can be used interchangeably as both are derived from the same tissue, succeeded by calcified masses, which are laid down in the form of cemental layers and bony trabaculae. As this process continues, the calcified tissues grow larger and fuse to form a dense sclerotic mass. Both cemental and osseous terms can be used interchangeably as both are derived from mesenchymal cells that have lost their ability to maintain their structural and functional morphology and, therefore, produce morphologically imperfect bone or cementum.

To elaborate, the mechanism involves localized areas of bone being replaced by fibrous tissue, succeeded by calcified masses, which are laid down in the form of cemental layers and bony trabaculae. As this process continues, the calcified tissues grow larger and fuse to form a dense sclerotic mass. Both cemental and osseous terms can be used interchangeably as both are derived from mesenchymal cells that have lost their ability to maintain their structural morphology and thus producing dysplastic bone. These cells are neither neoplastic nor premalignant.

Florid cemento-osseous dysplasias represents symmetric lesions affecting sextants and sometimes all quadrants may be involved but the predilection for posterior region of mandible is 78%. Florid cemento-osseous dysplasia has predilection for black females, which may suggest that this dysplastic process is related to hormonal imbalance that influences bone remodeling during fourth to fifth decade of life. It is more prevalent in blacks (78%) than in whites (5%) and Asians (4%). Among Asians, Indians form 2%. Out of the 11 cases reported so far, 10 were females and one was a 14-year-old male with involvement of all the quadrants. Pain is not a common finding in florid cemento-osseous dysplasia, only 4 patients (38-year-old female, 60-year-old female, 36-year-old female and 41-year-old female) out of total 11 cases reported in India had pain associated with the lesion [Table 2].

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Cemento-osseous dysplasias also show mixed radiolucent radiopaque masses associated with tooth bearing regions of the jaws. Multiple lesions may be seen in florid cemento-osseous dysplasia involving two to four quadrants. But, florid cemento-osseous dysplasias have a tendency to occur in black females. Histopathologically, they show the presence of irregular trabeculae of immature woven bone or osteoid, multinucleated giant cells and also, the presence of dysmorphic cementum or cementoid with basophilic reversal lines. The present case showed histopathological features suggestive of cemento-osseous dysplasia along with multiquadrant involvement with well-defined radio-opaque lesions. Thus, it was diagnosed as florid cemento-osseous dysplasia, though the age and sex parameters were not suggestive of this lesion. These lesions are usually asymptomatic, but our patient presented with mild discomfort, which was probably due to mandibular nerve impingement.
Table 2: Clinical features of cases of florid cemento-osseous dysplasia reported in the Indian population

| Authors          | Age/sex | Site                              | Pain     |
|------------------|---------|-----------------------------------|----------|
| Barbara et al.   | 60/female | Posterior mandible bilateral      | Present  |
| Sanjai et al.    | 20/female | Maxilla and mandible              | Absent   |
| Mangla et al.    | 30/female | Posterior mandible bilateral      | Absent   |
| Mangla et al.    | 38/female | Posterior mandible bilateral      | Present  |
| Jerjes et al.    | 27/female | Maxilla and mandible              | Absent   |
| Bansal et al.    | 14/male  | Maxilla and mandible              | Absent   |
| Kannan et al.    | 56/female | Posterior mandible bilateral      | Present  |
| Kannan et al.    | 47/female | Posterior mandible bilateral      | Absent   |
| Rao et al.       | 41/female | Posterior mandible bilateral      | Absent   |
| Asnani et al.    | 63/female | Posterior maxilla bilateral       | Absent   |
| Premalatha et al.| 37/female | Male bilateral                    | Absent   |

*osteofibrosis dysplasia in tibia and fibula only

CONCLUSION

Florid cemento-osseous dysplasias are benign, non-neoplastic, asymptomatic, self-limiting lesions. However, exposure of the sclerotic masses to the oral cavity may lead to secondary infection, which may be difficult and complicated to manage. Since the radiographic and histologic spectrum is similar to other fibro-osseous lesions with a different treatment plan, an accurate diagnosis is important. Surgical intervention is not the choice of treatment, as it may result in secondary complications; hence, patient is kept under observation and frequent follow-ups.

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