Fibro-osseous lesions of jaws: Analysis of three cases

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ABSTRACT

Fibro-osseous lesions (FOL) represent a varied group of entities in which the normal bone is replaced by cellular fibrous tissue containing mineralized foci that may vary in amount and appearance. These group of lesions predominantly affects the jaws and craniofacial bones. According to Waldron 1993, FOL includes three major groups namely fibrous dysplasia, cemento-osseous dysplasia, and ossifying fibroma. FOL show considerable overlapping in the clinical, radiographic and histological features, and so a thorough knowledge of these lesions is mandatory for interpretation and appropriate diagnosis. This article documents the clinical, radiographic and histological features of three cases of FOL and discusses the considerations related to diagnosis.

Key words

Cemento-ossifying fibroma, fibro-osseous lesion, fibrous dysplasia

INTRODUCTION

The term Fibro-osseous lesion (FOL) is a generic designation of a group of jaw disorders characterized by the replacement of bone by a benign connective tissue matrix. Conventional ossifying fibroma (COF), juvenile psammomatoid ossifying fibroma (JPOF), juvenile trabecular ossifying fibroma (JTOF), fibrous dysplasia (FD), and cemento-osseous dysplasia (COD) all belong to the fibro-osseous group of lesions. JPOF, JTOF, and COF are considered as true neoplastic lesions, while FD and COD are the developmental and reactive lesions, respectively.

CASE REPORTS

Case 1

A 24-year-old female presented with a slowly progressive painless swelling over right mandibular body since 10 years. There was no relevant medical and dental history. On inspection cafe-au-lait pigmentation of size

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10 cm × 10 cm approximately was present at the left thoracic region extending superiorly up to the clavicle. Extraoral examination showed a solitary swelling, approximately of 4 cm × 3 cm size over the right mandibular body which was bony hard and nontender on palpation. The overlying skin was intact and no parasthesia was noted. Intra oral examination revealed a localized, nontender, bony hard swelling extending from teeth 42 to 46 with buccal cortical plate expansion obliterating the buccal vestibule [Figure 1]. Based on the history and clinical findings a provisional diagnosis of benign FOL probably FD was given, and differential diagnosis included were COF, focal COD, JOF, Brown's tumor.

Vitality test revealed no response in relation to teeth 46, 47. Routine hemogram and serum calcium, phosphorous, and alkaline phosphatase were within normal limits.

The Intraoral periapical radiograph revealed an ill-defined radiopacity involving the periapex of teeth 43–45 with a displacement of 44, 45. Mandibular cross-sectional occlusal view revealed a radiopacity with orange peel appearance extending from 43 to 46 and definite bucco-lingual cortical expansion was evident [Figure 2]. Orthopantomogram revealed a radiopacity extending from 43 to 46 with a displacement of 44, 45 and altered trabecular pattern. Axial computed tomographic (CT) revealed a solitary, hyperdense lesion with thinning of buccal and lingual cortices [Figure 2]. Chest X-ray and radiographs of other long bones revealed a normal

study. Histopathologic examination revealed a fibrous connective tissue stroma with irregularly shaped trabeculae [Figure 3].

Case 2

A 36-year-old female presented with a slowly progressive painless swelling in the chin region since 2 years. There was no significant medical and dental history. Extra oral examination revealed mild facial asymmetry due to a swelling in anterior mandible along right side measuring 2.5 cm × 2.5 cm size approximately which was bony hard and nontender with no parasthesia. Intra oral examination revealed a well-defined swelling in relation to teeth 42, 43, 44 obliterating the labial vestibule and was bony hard, nontender with intact overlying mucosa. Significant buccal cortical expansion was noted from 41 to 44 and lingual expansion from 42 to 44 [Figure 4]. Correlating the history and clinical findings a provisional diagnosis of benign FOL was given. Differentials considered were ameloblastoma, odontogenic myxoma,



Figure 1: Intraoral swelling extending from teeth 42 to 46 with buccal expansion

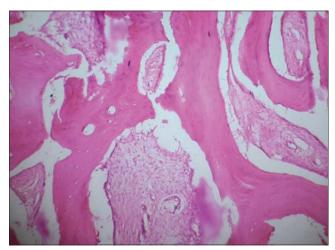


Figure 3: Histological picture showing immature woven bone with fibrous

calcifying epithelial odontogenic tumor (CEOT), central giant cell granuloma, and central odontogenic fibroma.

In vitality test, there was a delayed response in tooth 42 and lesional aspiration was nonproductive. Routine hemogram and serum chemistry were within normal limits. The occlusal radiograph revealed a mixed radiopaque - radiolucent lesion extending from 34 to 45 buccally, and 36 to 46 lingually with significant bucco-lingual cortical expansion and ground glass appearance was noted [Figure 5]. Orthopantamogram revealed a well-defined mixed radiopaque - radiolucent lesion involving periapex of teeth 43 to 45. Axial CT showed a heterodense expansile mass with the thinning of buccal and lingual cortices [Figure 5]. Histopathologic examination revealed a fibrous stroma with ill-defined trabeculae and osteoblastic rimming suggestive of FD [Figure 6].

Case 3

A 40-year-old woman reported with a swelling in the chin region for the past 3 months which was progressively increasing in size. The medical and dental histories were not significant. Extra orally, a significant facial asymmetry was noted due to a swelling of size approximately 4 cm × 5 cm in relation to the anterior



Figure 2: Occlusal view revealing diffuse radiopacity with orange peel appearance and axial computed tomographic image showing a hyperdense lesion with bucco-lingual cortical expansion



 $\textbf{Figure 4:} \ Intra \ or al \ swelling \ from \ 42 \ to \ 44 \ with \ obliteration \ of \ labial \ vestibule$

mandible which was bony hard and nontender. Intraoral examination revealed a well-defined swelling on the labial aspect approximately 4 cm × 6 cm size extending from the teeth 43 to 35 crossing the midline with obliteration of labial vestibule and a swelling lingually extending from 31 to 34 with the mild displacement of the lower anterior teeth. The swelling was firm to hard in consistency with bucco-lingual cortical expansion and intact overlying mucosa [Figure 7]. A provisional diagnosis of benign odontogenic tumor probably ameloblastoma was given and differential diagnosis included variants of ameloblastoma, COF, COD, central giant cell granuloma, CEOT, and hyperparathyroidism.

Thermal vitality test revealed no response in relation to tooth 34 and lesional aspiration was nonproductive. Routine hemogram was within normal limits.

The Intraoral periapical radiograph revealed a radioluceny from the periapex of teeth 41 to 33 and root resorption in relation to 33. The anterior mandibular occlusal view showed buccal cortical expansion extending from 41 to 37. A panoramic radiograph revealed a well-defined radioluceny with radiopaque foci extending from the teeth 42 to 36. Axial CT revealed a heterogenous osteolytic lesion crossing the midline with the expansion of bucco-lingual cortices [Figure 8]. Incisional biopsy reported a fibrous connective tissue stroma with eosinophilic and basophilic

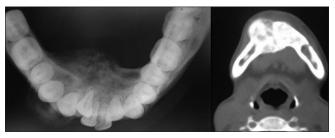


Figure 5: Occlusal radiograph showing ground glass appearance and axial computed tomographic image depicting a heterodense mass with thinning of buccal and lingual cortices



Figure 7: Intra oral photograph depicting bicortical expansion

calcified areas along with cementicle like substances suggestive of cemento-ossifying fibroma [Figure 9].

DISCUSSION

FD is a nonhereditary, developmental condition occurring predominantly within the long bones and craniofacial skeleton. The most common location within the craniofacial complex is the maxilla, where it presents as a painless, slow growing diffuse lesion in children and young adults. FD can occur as monostotic or polyostotic forms, with or without a syndromic association. The monostotic type accounts for 80% of cases seen. There is an equal gender distribution, but polyostotic forms are more common in females. [4,5] In case 1 and case 2, the lesion occurred in the mandible involving the molar-premolar region and both the cases belonged to monostotic type. FD has its onset usually in late childhood or early adolescence which was in coincidence with both the cases. The slowly progressive enlargement of the affected jaw is usually painless and typically presents as a unilateral swelling. As the lesion grows, facial asymmetry becomes evident and may be the initial presenting complaint. [6] In case 1 and case, 2 the presenting complaint was facial asymmetry which was asymptomatic with a unilateral presentation. Radiographically, FD displays an abnormal opacification which ranges from very numerous, small,

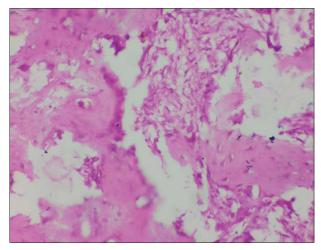


Figure 6: Histological picture showing fibrous connective tissue stroma with osteoblastic rimming



Figure 8: Orthopantamogram showing mixed radiolucent - radioopaque lesion and axial computed tomographic image revealing an expansile lesion with bulging of cortical bone

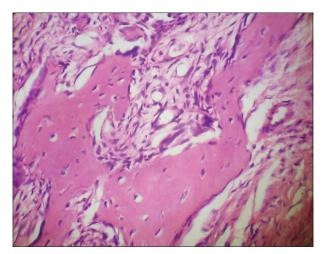


Figure 9: Histopathological picture showing fibrous connective tissue stroma with calcified areas and cementicle like substances

and diffusely distributed opacities ("ground glass" and "peau-d'orange") to sclerosis, classically described as "cotton wool."[7] Case 1 revealed a radiopacity depicting an orange peel appearance and case 2 manifested a ground glass appearance. Histology of FD consists of fibrous connective tissue proliferation containing irregularly shaped trabeculae of immature bone. The trabeculae assume bizarre shapes linked to Chinese character[8] which was noted in case 2. Histology of case 1 showed lamellated trabeculae with osteoblastic rimming and minimal fibrous tissue components resembling COF, but the radiographs depicted a homogenous radiopacity with no clear demarcation from the surrounding bone which was in favor of FD. Correlating the clinical, radiographic and histological findings the final diagnosis of case 1 and 2 was solitary monostotic FD. The main differential diagnosis of FD is COF. FD presents as a poorly differentiated lesion, diffuse and blends with the surrounding bone with characteristic ground glass appearance whereas COF are usually well defined, and they occasionally have a soft-tissue capsule. [9] Waldron[6] reported cases of FD showing lamellated trabeculae and osteoblastic rimming which may confound diagnosis as they resemble COF. Other entities which may be confused with FD are COD, Pagets disease, cementoma, cherubism, hyperparathyroidism, chronic sclerosing osteomyelitis, and osteogenic sarcoma, etc. [10]

Cemento-ossifying fibroma is a benign nonodontogenic tumor that contains multipotential cells that are capable of forming cementum, lamellar bone, and fibrous tissue. Most frequently occurs in females with an age range of 10–59 years^[11] coinciding with case 3 where the patient was a 40-year-old female. Around 62–89% of the tumor arises in the mandible with 72% occurring in the premolar region^[12] which is in contrast to the present case where it occurred in the anterior mandible. The possible etiopathogenesis may be a tooth extraction, history of previous trauma, generalized periodontitis, and genetic factors. In our case, the patient had generalized

chronic periodontitis which might be an etiological factor. While one-half of all cases is asymptomatic, the growth of the tumor over time may lead to facial asymmetry, with the mass causing mandibular expansion and possible displacement of dental roots.[13,14] Our case had a significant facial asymmetry, migration of the lower anterior teeth and bicortical expansion. Radiographically, the tumor appears as a radiolucent lesion with no internal radiopacity in its early stages. As the lesion matures, there are increasing calcific flecks progressing to a radio opaque mass. [15] This case manifested as a well circumscribed radiolucent lesion with internal radiopacities and root resorption was also noted. CT image revealed an osteolytic lesion with the expansion of bucco-lingual cortices and there was no cortical violation. In its histopathology, it is typical to encounter a benign fibroblastic stroma with mineralized tissue masses of basophilic aspect which correspond to osteoid or cementoid material[13] concurring with the present case. Differential diagnosis includes FD, COD, CEOT, and a calcifying epithelial odontogenic cyst. COD is usually multifocal and present with a wide sclerotic border where as Pindborg's tumor has a high association with an impacted tooth.[9]

CONCLUSION

The diagnostic criteria of FOL are contentious and it is often difficult to distinguish these lesions from one another due to the striking similarities in the clinical, radiological, and histological findings. So meticulous observation of the patient's history, age, sex, site of the lesion, the internal architecture of the radiographs, and histological findings may provide the key to the otherwise challenging diagnosis.

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