

CASE REPORT



Focal osseous dysplasia with an unusual and rare clinical feature of numbness

Ishita Agarwal, Ranjani Shetty, G.P. Sujatha, L. Ashok

Department of Oral Medicine & Radiology, Bapuji Dental College and Hospital, Davangere, Karnataka, India

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Correspondence

Dr. Ishita Agarwal, Department of Oral Medicine & Radiology, Bapuji Dental College and Hospital, Davangere - 577 004, Karnataka, India. Phone: +91-7406051273, Email: vanikagupta@gmail.com

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Abstract

Focal osseous dysplasia (FocOD) is a non-neoplastic benign fibro-osseous lesion. Osseous dysplasias (OD) are commonly seen as incidental finding on adult dental radiographs. This entity occurs most commonly in females and is almost always asymptomatic and non-expanding. This case report presents a case of FocOD in the posterior mandible with an unusual clinical feature of numbness of right lower third of the face in a 45-year-old male patient, which was confirmed by histopathological report. Radiological, clinical, and histopathological characteristics of the FocOD and similar lesions are discussed.

Introduction

Osseous dysplasia is a reactionary and non-neoplastic lesion, which usually presents as asymptomatic lesion in tooth-bearing areas of the jaw. In previous versions of the WHO classification of fibro osseous lesions,^[1,2] these were referred to as cemento-osseous lesions. They were clearly divided into three entities, periapical cement-osseous dysplasia, Focal cement-osseous dysplasia (FocOD), florid cement-osseous dysplasia (FCOD). In the recent classifications^[3] they have been termed as osseous dysplasias (OD). Focal osseous dysplasia (FocOD), periapical osseous dysplasia (POD) and florid osseous dysplasia (FOD).^[4] It is now appreciated that they represent a spectrum of lesions, probably reactive in nature, which only differ by their clinical presentation and radiological appearances. All types affect the tooth-bearing areas of the jaws and appear to arise from the periodontal ligament and form cementum, or cementum-like tissue. For unknown reasons, they all have a predilection for black females in the third to fifth decades, where the prevalence may be as high as 6%.^[1,5] FocOD presents as a solitary lesion in the posterior jaw, most often the mandible, and usually at the site of a previous tooth extraction. Occasionally, they may be found at the apices of a molar tooth. The exact etiology and pathogenesis of FocOD is not known. FocCOD/FOD is diagnosed on the basis of clinical, radiographical and histological features.

Case Report

History

A 45-year-old male patient came to Department of Oral Medicine and Radiology, Bapuji Dental College and Hospital, with a chief complaint of pain in right lower back tooth region since 3 days and reduced sensation on right lower third region of the face since 10 years. The history of present illness revealed that the numbness was present since 10 years, which started after the extraction of a tooth on the same side and had started as a moderate loss of sensation, which the then dentist had told would subside gradually, associated history of mild pain since 10 years, was gradual in onset, intermittent in nature, mild to moderate in intensity, which had increased in intensity since 2-3 days, no aggravating and relieving factors were present. He gave a past dental history of extraction of right mandibular first and third molar and left mandibular first molar 10 years back due to caries and followed by fabrication of a fixed prosthesis 10 years back. Past medical history was not significant. Patient had a family history of diabetes in both mother and father and had no habit history.

Extra and intraoral examination

On general physical examination, the patient was moderately built and nourished, all the vital signs were within the normal

limits. On intraoral examination, soft tissue examination revealed generalized mild gingival recession, with mild gingival bleeding on probing. On Hard tissue examination 18, 36, 46, 48 were clinically missing, amalgam restoration was present in relation to 15 and 17. Fixed partial denture in relation to 35 to 37 and 45 to 47. Slight expansion of the buccal and lingual cortical plate in relation to 45 to 47 [Figure 1] was present, which was tender on palpation. 47 was tender on percussion. Test for parasthesia was done and was compared on both the sides, where patient felt reduced sensation on the right side of mandibular attached gingiva, labial mucosa and lip compared to the left.

Differential diagnosis

Considering the chief complaint and history a provisional diagnosis of a benign cyst or tumor pressing over the nerve was considered and a clinical differential diagnosis of Odontogenic cyst like residual cyst, since it has a predilection for adult male and occurs commonly in posterior mandible, and usually associated with a history of extraction of the tooth; tumors like ameloblastoma was considered since it commonly occurs in the mandibular body, with cortical plate expansion and associated paresthesia; nerve sheath tumors like neurolimoma was also considered, however, intraoral lesions are rare, but if it occurs then mandible is most common site and occurs in second to third decade of life, it can present with swelling or bone expansion with pain and paresthesia, which was present in our patient also, and rarer entities like metastatic malignancy was considered since, it also occurs in a molar area of mandible and mimic a reactive or benign lesion which can invade bone and nerve leading to paresthesia.

Investigations

The patient was subjected to the routine investigations such as complete hemogram, random blood sugar and Hb1ac, which was within the normal limits, OPG was taken as a screening radiograph, IOPA, Mandibular occlusal-right lateral topographic view, Mandibular right lateral oblique body view were done. OPG [Figure 2] revealed a well-defined mixed radiolucent radioopacity in relation to 47 with non-sclerotic margins, measuring about 3 cm × 3 cm, round to oval in shape extending from distal root of 47 to 2 cm anterior to angle of mandible anteroposteriorly, and superoinferiorly extending from middle third of the distal root of 47 to the inferior border of mandible. Internal structure showed mixed radiolucent radioopacity, effect on the surrounding structures was not very evident with loss of lamina dura in the apical one-third of the distal root of 47 and no effect on the inferior alveolar nerve canal radiographically. IOPA [Figure 3] revealed loss of lamina dura in the apical one-third of the distal root of 47 and radiolucency in the furcation area and moderate horizontal bone loss. Considering these, features a radiographic differential diagnosis of ossifying fibroma and FocOD in relation to 47 were thought of.

Final diagnosis

Incisional biopsy was done, histopathological report revealed mature bony trabeculae and small focus of subperiosteal fibrosis



Figure 1: Intra oral photograph showing fixed prosthesis in relation to 45, 46, and 47. Mild buccal and lingual expansion present in relation to 47



Figure 2: Orthopantomogram showing missing 46, and mixed radiolucent and radioopaque lesion in posterior body of mandible on right side



Figure 3: Intra oral periapical radiograph in relation to 47 showing internal structure of mixed radiolucent and radioopaque structure present in relation to 47

with immature bone formation. In view of the foci of fibrosis with immature bone and periapical location of the lesion, a final diagnosis was given as FocOD in relation to 47. Complete excision and curettage were done. After 6 months the patient was recalled and reviewed, the paresthesia had reduced by 50% and the radiograph showed normal bone formation, the patient is kept under follow-up. After 1-year, patient was again reviewed, and there was complete resolution of paresthesia with mild tingling sensation was present.

Discussion

Fibro-osseous lesions “represent a diverse group of reactive, dysplastic and neoplastic proliferations characterized by replacement of normal bone with a collagenous matrix containing trabeculae of immature bone and in some instances psammomatous deposits frequently described as “cementum-like material.”^[6]

OD are variants of non-neoplastic fibro-osseous lesions.^[7] FocOD represents the most common fibro-osseous lesions of the jaws. These benign lesions are characterized by an alteration of bone structure. The normal architecture of bone is replaced by fibroblasts and collagen fibers, and also by a variable quantity of mineralized material.^[7] Previously, Waldron, observing its localized nature, first reported it as the “localized fibro-osseous-cemental lesion,” which Summerlin and Tomichre named as focal cement osseous dysplasia. While the exact etiology of FocOD is unclear, they are favored to arise from medullary bone and/or periodontal ligament tissue. Hormonal imbalances affecting bone remodeling are currently being investigated.^[8]

FocOD occurs predominantly in females with a mean age in the mid-thirties, and a slightly higher distribution among African Americans (64%). The majority of these lesions are asymptomatic (62%) with an average size of 1.5 cm.^[9] When symptomatic, the lesions most commonly cause pain and swelling. Waldron and co-authors claimed that most of their symptomatic cases had been “edentulous in the affected areas for many years.”^[8] Occasionally, FocOD can lead to secondary pulpal and periodontal infection and progress to necrosis.^[10] The mandible is the most frequent site of occurrences (86%). Secondly, it shows a close association with tooth apices (70.6%) or with previous extraction sites (21%).^[9] Ohkura reported that two-thirds of the edentulous FocODs occurred in the posterior mandible. Since so many FocODs occur at extraction sites, they may partly represent as suggested by Waldron, the end-stage of an abnormal reaction of bone to injury.^[8]

The condition rarely produces expansion of the bone. Larger lesions may however cause slight jaw enlargement.^[11] The paresthesia noticed in the above discussed case may be due to the increased size of the lesion causing changes in the nerve on which it was impinging for a long duration.

Initially, the paresthesia was thought to be due to injury to the nerve during previous extraction but once the patient came back with reduced numbness after curettage of the lesion

it was considered to be due to the FocOD though it is a rare clinical feature. The latest classification of osseous dysplasia includes the concept of spectrum of clinicopathological entities in which the diagnosis can only be made on the basis of a full consideration of clinical, radiological and histological features. This classification includes, POD, which involves a small number of teeth, and it's localized only in the anterior region of the mandible, FocOD which is the same lesion as POD, but localized in the posterior region; FOD is a more extensive lesion that may be confined to two quadrants or more; and familial gigantiform cementoma, which involves multiple quadrants, while being expansible, and it shows an autosomal dominant inheritance.^[12]

FocOD has three developmental staging, each with specific radiographic features. Early or osteolytic stage, in this stage radiographs, revealed well-defined radiolucency with loss of lamina dura, intermediate and cementoblastic stage in which opaque areas are present within the radiolucency, display radiographic picture of mixed radiolucent and radiopaque lesion, last osteosclerotic or inactive stage, in which definite radioopacity is present and only at this stage well defined border is seen as radiolucent line.^[13]

Histopathologically, FocOD consists of a cellular and fibrous connective tissue stroma, which is benign, heterogeneous and punctuated by irregular osseous (mature and immature bone) and/or cementum-like calcifications [Figure 4].^[4]

The radiographic differential diagnosis includes periapical granuloma or cyst and chronic osteomyelitis in the osteolytic stage, in the mixed and radioopaque stages, chronic sclerosing osteomyelitis, ossifying/cementifying fibroma, odontoma and osteoblastoma can be considered according to the development.^[14]

In our case, osseous dysplasia was considered as, osseous dysplasia will be round to oval in shape, predominantly display mixed radiolucency and radioopacity with an ill-defined radiographic border, whereas mature lesions are usually uniformly dense with thin radiolucent border and root resorption usually is absent. Moreover, osseous dysplasia commonly found to be associated with periapex or previous extraction site, as was in our case.

Ossifying fibroma was also considered since, it presents as well-defined radiolucency with or without sclerotic borders or as a mixture of radiolucency and radioopacity, which is second

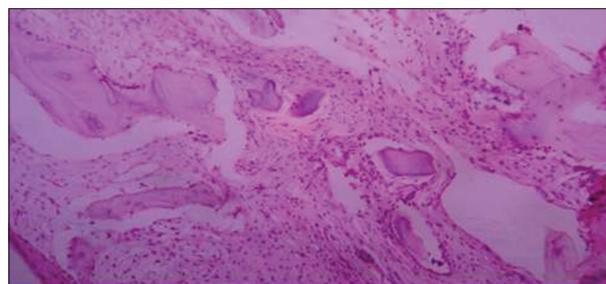


Figure 4: Highly cellular fibrous connective tissue, showing focal areas of ossification

most common appearance with variable degree of jaw expansion. However, ossifying fibroma will almost always have well-defined border which helps in differentiating with osseous dysplasia radiographically.

FocODs are generally symptomless and need no treatment; their clinical importance has been emphasized by their presence in edentulous sites required for osseointegrated implants. As an essential prelude to its management in this regard, a better understanding of the frequency and presentation of the FocOD in the global literature is required.^[8] To confirm the diagnosis follow-up is a must. Biopsy is recommended when lesions become symptomatic, show radiological features similar to ossifying fibroma, or occur at a site that will be used for an implant.^[10] Recall visits are enviable because according to one school of thought there are the possibility of transformation of FocOD into FOD.^[14]

Conclusion

FocOD represents one of the most common fibro-osseous lesions of the jaws, which is often symptomless and presents as an incidental radiographic finding. The above case having a unique feature of associated numbness and mild pain is a rare finding, which concludes that paresthesia can be one of the rare clinical features of FocOD.

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