Cone Beam Computed Tomography Findings in Fibrous Dysplasia of Maxilla: A Case Report

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ABSTRACT

Introduction: Fibrous dysplasia (FD) is a benign fibro-osseous disease which causes replacement of normal bone by fibrous connective tissue and immature bone, usually affecting the children and young adults. It can be monostotic or polyostotic, affecting maxilla twice as compared to mandible. FD may cause facial deformities, bone pain & pathologic fractures in advanced cases.

Case report: The present case report describes FD in 16 year old female patient presented with swelling on the right side of the maxillary arch highlighting the CBCT features.

Conclusion: Conventional radiography makes it difficult to delineate the exact extension of the lesion, due to complex facial anatomy so Cone Beam Computed Tomography (CBCT) should be the method of imaging for evaluating this lesion and its relationship with adjacent structures.

Keywords: CBCT, Diagnosis, Fibrous Dysplasia

INTRODUCTION

Fibrous dysplasia (FD) is a genetically-based sporadic disease of the bone, and it constitutes 2.5% of all osseous & 7% of all benign bone tumors. 1,2 Mutations in the gene GNAS 1, encoding for the α subunit of a signal transducing G protein (Gs-a) causes proliferation and differentiation of preosteoblasts which is responsible for development of this lesion. 1

FD presents in three forms – monostotic (affects only one bone), polyostotic (affects multiple bones) and craniofacial. The monostotic form is most common and accounts for 73% of cases, 13% of cases are polyostotic, and remaining 13% are craniofacial cases.³ FD causes asymptomatic enlargement of involved bone which leads to facial asymmetry, associated with dental issues, such as prolonged retention of deciduous teeth and malalignment, thus affecting aesthetics, mastication and speech.^{4,5}

In our case, FD affecting the right side of maxilla causing bone expansion is presented along with the importance of CBCT imaging for diagnosis of this lesion.

CASE REPORT

A 16 year old female patient reported to the department of Oral Medicine & Radiology with the chief complaint of swelling on right side of maxilla since 5 years, swelling was gradually increased in size to the present size.

On extraoral examination there was a mild diffuse swelling

noted over right side of the mid face. The swelling was bony hard on palpation with normal overlying skin and temperature. Intraorally the swelling was extending from distal aspect of right maxillary canine (13) to second molar (17) region with a normal overlying mucosa. Buccal expansion was more prominent as compared to palatal expansion. Obliteration of buccal vestibule and displacement of second premolar (15) was noted [Fig 1]. IOPA revealed loss of normal trabecular pattern resulting in a ground glass appearance in premolar and molar region. OPG revealed developed 11, 12, 13, 14, 15, 16, 17, 21, 22, 23, 24, 25, 26, 27, 31, 32, 33, 34, 35, 36, 37, 41, 42, 43, 44, 45, 46, 47 and developing 18, 28, 38, 48. There is a uniformly radiopaque lesion noted in right maxilla extending from distal aspect of 13 to 18 anteroposteriorly, and from infraorbital margin to alveolar crestal region superoinferiorly showing a ground glass appearance. Enostosis was noted at periapical region of 45 [Fig 2].

CBCT interpretation

CBCT examination was performed using CS9300 scanner with acquisition protocol as follows: 85 kVp, 8 mA, 11.26 seconds and field of view (FOV) of 17x13. Multiplanar reconstruction was done and images were acquired in axial, coronal, sagittal and three-dimensional views [Fig 3, 4, 5].

A well defined hyperdense lesion noted in right maxilla extending from 13 to 18 anteroposteriorly, infraorbital



Figure-1: Intraorally the swelling extending from distal aspect of right maxillary canine to second molar.



Figure-2: OPG revealed changes in normal trabecular pattern in right maxilla.

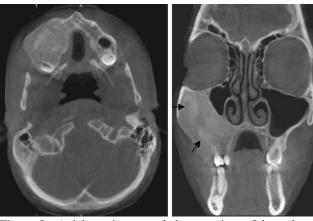
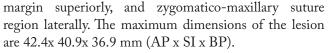


Figure-3: Axial section revealed expansion of buccal and palatal cortices on right side; **Figure-4:** Coronal section revealed reduction in volume of right maxillary sinus without any change in shape. Also areas of varied density are appreciated within the lesion



The periphery of the lesion is partly corticated, and the internal of lesion is hyperdense with areas of varied density (heterodense). There is expansion of buccal and palatal cortices, along with displacement of walls of maxillary sinus resulting in reduction of the volume of sinus, but the overall shape of the maxillary sinus is maintained.

Palatal displacement of 15, changes in the lamina dura and periodontal ligament was noted but without any root resorption.

Additional findings: Mild deviation of nasal septum towards right side and hypertrophy of inferior nasal chonchae was noted on left side. There was a small well defined hyperdense mass noted at the pericaical region of 45 suggestive of enostosis.

DISCUSSION

Fibrous dysplasia was first described by Lichtenstein in 1938. Jones described hereditary familial form of localized FD which is called as cherubism. ⁴ There are three forms of this entity, monostotic, polyostotic and craniofacial. Three

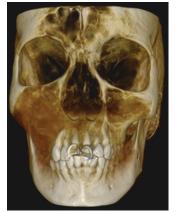


Figure-5: 3D Reconstruction.

percent of the polyostotic form have endocrinopathies and are cases of McCune–Albright syndrome.¹

The term monostotic can be used for cases involving mandible, but it should not be used for cases of FD affecting maxilla because there it can involve adjoining bones such as the zygoma, and then it will be categorized under craniofacial FD. ¹

FD is usually found in young adults, as seen in our case of a 16 year old female patient. The lesion becomes static when skeletal growth ceases, but may become active again in cases of pregnancy, use of oral contraceptives and early surgical intervention. There is no gender predilection for fibrous dysplasia except for McCune Albright syndrome which affects females predominantly. Maxilla is more commonly involved than mandible and frequently in posterior aspects of bone.

The usual presentation is a painless swelling involving the jaw bones, which may cause deformity of the face. There can be displacement of the teeth but the teeth usually remains firm. ^{6,7} The complications involve pain and pathologic fractures, which are rare. The fifth nerve impairment, hearing loss, anosmia and seizure disorders have been reported as neurological impediments. ⁴

The radiographic appearance of FD depends on the stage of development and amount of bony and fibrous matrix within the lesion.⁸ Early lesions appear radiolucent, and mature lesions may appear sclerotic.⁹ The different radiographic patterns on the basis of radiodensity are ground glass appearance, orange peel appearance, cottonwool appearance, amorphous dense and fingerprint pattern.⁶ The most common radiographic appearance is ground glass opacification which results from superimposition of numerous poorly-calcified bony trabeculae arranged in a unsystematic pattern as in present case.⁸

Lesions of FD usually present with ill-defined borders and gradual blending with normal bone on plain radiography while 3D assessment with CBCT displays well-defined corticated lesions with sharp borders. This well-defined radiographic appearance results from precise evaluation of the affected bone with CBCT opposed to plain radiography as noted in present case.⁸

Differential diagnosis of FD in the early stage includes, central ossifying fibroma, central giant cell granuloma and aneurysmal bone cyst. In late stages it should be differentiated from Paget's disease and osteomyelitis.³

The factors guiding the approach of treatment involves, age of patient, site of lesion and the intensity of the symptoms. The FD lesion is benign in nature so the surgery should be relatively conservative with the key intention of preserving the function. There are reports of use of Calcitonin & Pamindronate in the treatment of FD.²

Radiotherapy is contraindicated as the lesion is radioresistant and there are increased chances of sarcomatous transformation of the affected bone after the radiation.¹⁰

Lesions of FD generally get stabilized after puberty and that's why our patient was instructed to undergo regular imaging examinations so the evolution of lesion could be followed till skeletal maturity.

CONCLUSION

Fibrous dysplasia is an entity which should be considered in the differential diagnosis of young patients reporting with unilateral facial swelling. Radiographic evaluation is must as presentation and distribution of the lesions may define the diagnosis. CBCT plays a important role in assessment of lesion and determining the exact extent of the lesion including pre-surgical evaluation and follow-up of the patients.

The increased expertise of the imaging characteristics of fibrous dysplasia can help the dentist in the early diagnosis and management of patients.

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