



Case Report

Fibrous dysplasia of the anterior mandible: A rare case report

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Received : 02-Sep-2017
Revised : 24-Oct-2017
Accepted : 09-Nov-2017

ABSTRACT

Fibrous dysplasia (FD) is a rare bony disorder in which normal bone is replaced by abnormal fibro-osseous tissue. It often involves the long bones, craniofacial bones, ribs, and pelvis. Approximately 30% of monostotic FD (MFD) lesions are found in the cranial or facial bones. In general, FD is found in teenagers, and it usually becomes static after adulthood. FD involves the maxilla almost two times more often than the mandible. It frequently appears in the posterior region of the jaw bone and is usually unilateral. Here, we present an unusual case of symptomatic MFD affecting the anterior region of the mandible in a 43-year-old female with the clinical, radiographical, and histopathological features. The clinical examination showed both the labial and lingual bone expansion in the anterior mandible. The radiographic examination revealed a lesion with both radiopaque and radiolucent features showing a “ground-glass” appearance. The diagnosis was obtained after confirmatory intrabony biopsy with the histopathological examination, and it was diagnosed with benign FD. The patient preferred regular follow-up of MFD after discussion. During the regular follow-up, MFD lesion showed no obvious signs of progression or malignancy features.

KEYWORDS: *Fibrous dysplasia, Mandible, Middle-aged person, Monostotic*

INTRODUCTION

In 1938, Lichtenstein first coined the term “fibrous dysplasia (FD)” [1]. FD is a rare, nonmalignant condition in which normal bone and marrow are replaced by fibrous tissue and randomly distributed woven bone, usually with pain, bony deformity, and pathologic fractures [2]. In general, FD presents in three forms – monostotic, polyostotic, and polyostotic with endocrinopathies, which can be associated with hyperpigmentation and endocrinological disorders and is called McCune–Albright syndrome [3]. Common sites of skeletal involvement are the long bones, craniofacial bones, ribs, and pelvis [2]. Monostotic FD (MFD), although less serious than polyostotic FD (PFD), is of greater concern to the dentist because of the relatively high frequency of occurrence in the jaws [4]. FD is caused by somatic activating mutations of the gene *GNAS* in a subunit of the stimulatory G protein, located at 20q13.2-13.3 [2]. The diagnosis of FD is based on physical, radiological, and histopathological examination. There are different treatment approaches including observation, medical treatment, and surgical treatment. This article presents a case of MFD in the anterior mandible in a 43-year-old female with the clinical, radiographical, and histopathological features.

CASE REPORT

A 43-year-old female presented at the Dentistry Department of Hualien Buddhist Tzu Chi Hospital with pain over the left

posterior teeth for a week. Extraoral examination showed no obvious facial asymmetry. However, intraoral examination showed both labial and lingual bone expansion in the anterior mandible. The labial expansion was located at teeth 22 and 23, and the lingual expansion was from tooth 22 to tooth 27 [Figure 1]. Radiographic examination revealed a lesion with both radiopaque and radiolucent features showing a “ground-glass” appearance from the mesial side of the tooth 21 root apex to the distal side of the tooth 27 root apex, and the two other small solitary apical lesions of teeth 18 and 31 [Figures 2 and 3]. All nine teeth mentioned above responded normally to the electric pulp test. The cone-beam computed tomography (CS 9300, Carestream Health, Rochester, NY, USA) revealed a “ground-glass” appearance in the buccal and lingual bony expansion regions with relatively well-defined borders [Figure 4]. Endosteal scalloping was also noted. Thinning of the cortical bone was observed in the sagittal view labially and lingually [Figure 4a], while the homogeneous sclerotic bone was shown in the transverse view labially [Figure 4c]. A three-dimensional rendered image revealed labial bony expansion [Figure 4d]. A confirmatory

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How to cite this article: Yang HY, Su BC, Hwang MJ, Lee YP. Fibrous dysplasia of the anterior mandible: A rare case report. Tzu Chi Med J 2018;30:185-7.

Access this article online	
Quick Response Code: 	Website: www.tcmjmed.com
	DOI: 10.4103/tcmj.tcmj_57_18

intrabony biopsy was done [Figure 5]. Grossly, the specimens of lesion were grayish and elastic. Histologically, there was

FD characterized by irregularly shaped trabeculae of woven bone and lamellar bone in a cellular, loosely arranged fibrous stroma [Figure 6]. After pain relief, we explained the possible risks and recommended suitable treatment plans for the patient.



Figure 1: Clinical photograph showing expansion of both the labial and lingual cortical plates of the anterior mandible



Figure 2: Panoramic radiography reveals a mixed radiolucent and radiopaque lesion with a ground glass appearance in the anterior mandible which is crossing the midline

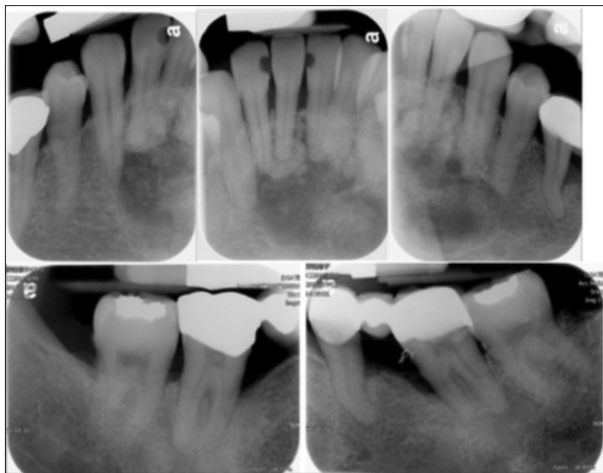


Figure 3: Periapical radiography demonstrates details of the mixed radiolucent and radiopaque lesion extending from the mesial side of the tooth 21 root apex to the distal side of the tooth 27 root apex and the two other apical lesions of teeth 18 and 31

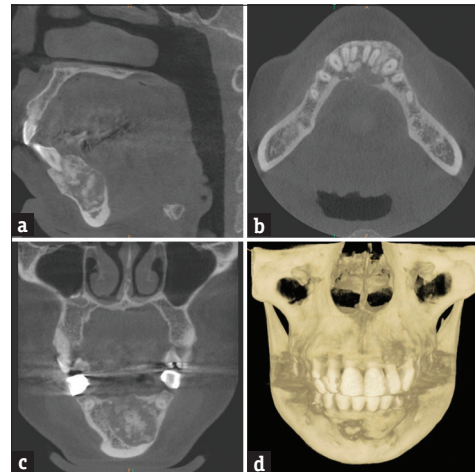


Figure 4: Cone-beam computed tomography reveals a "ground glass" appearance in the buccal and lingual bone expansion regions and relatively well-defined borders. (a) Sagittal view. (b) Coronal view. (c) Transverse view. (d) Three dimensional rendered image of the jaw bone



Figure 5: Biopsy performed at the most protruding site of the labial cortical plate

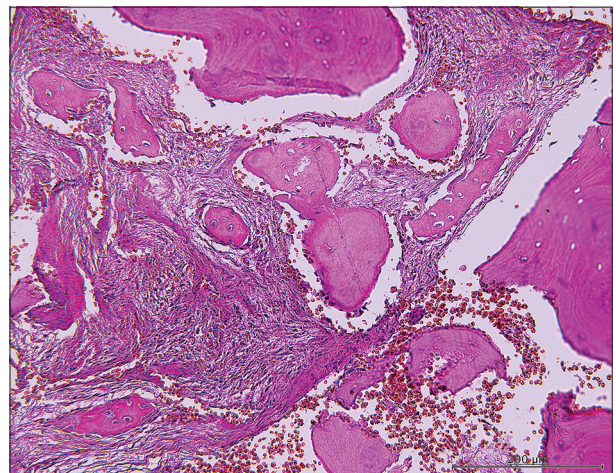


Figure 6: Irregularly shaped trabeculae of woven bone and lamellar bone in a cellular, loosely arranged fibrous stroma

The patient preferred noninvasive medication for pain relief and regular follow-up.

DISCUSSION

FD is a congenital, metabolic, nongenetic disturbance that represents 2.5% of all bone tumors and over 7% of all benign bone tumors [5]. FD involves the facial and cranial bones in nearly 50% of PFD patients and in 10%–27% of MFD patients [6]. The maxilla is more commonly involved than the mandible in MFD jawbone lesions. Our case involved the mandibular bone, this is a rare case in the FD. The lesion was in the anterior mandible crossing the midline with two other small lesions in the posterior region of the mandible. FD can occur at any age. However, it is usually observed in children and young adults with 75% of patients presenting before the age of 30 [2]. In one study reported that, FD has a female predilection and is observed more frequently in older decades [7]. In another study, no gender predilection is found [2]. After puberty or bone maturation, FD progresses slowly or ceases, whereas in another report, FD continued to progress into old age [8].

The differential diagnosis of FD includes simple bone cyst, nonossifying fibroma, osteofibrous dysplasia, adamantinoma, low-grade intramedullary osteosarcoma, and Paget's disease [2]. The current gold standard for the diagnosis of FD is a histologically-proven fibro-osseous lesion with poorly defined margins which are confirmed by radiographic findings [3]. Treatment protocols for FD include observation, medical treatment, and surgery. Clinical observation is suggested for FD lesions that have no risk of pathologic fracture or deformity [2]. Medical treatment with bisphosphonates may have benefits including improvement of function, pain relief, and lower fracture risk in appropriately selected FD patients [2]. One study reported clinical improvement in children and adults treated with bisphosphonates [9]. Surgery is indicated for confirmatory biopsy, correction of deformity, prevention of pathologic fracture, and/or elimination of symptomatic lesions [2]. Conservative management has been the standard of care, which involves removing the diseased bone via an intra-oral approach. Cortical bone grafts are superior to cancellous bone grafts or bone-graft substitutes because of the excellent quality of the remodeled cortical bone [2]. After a confirmatory biopsy, our case was diagnosed with symptomatic MFD. After discussion with the patient, she refused invasive surgical treatment and chose conservative management with regular recall and clinical observation. The reported prevalence of malignant transformation of FD is 0.4%–4% [2]. Therefore, periodic

follow-up, for example, every 6 months, and radiographic examination should be carried out to verify that there is no progression or malignancy [2]. MFD of the anterior mandible in the middle-aged female is rare. Nevertheless, the general dental clinician can be the first to detect this disease in daily practice using panoramic radiography. Therefore, sufficient knowledge of FD is essential to make an appropriate diagnosis and reduce the complications of the disease. With improvements in medicine and molecular technology, better therapy for FD, such as genetic therapy, may be possible in the near future.

Declaration of patient consent

The authors certify that the patient has obtained an appropriate patient consent form. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published, and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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