CASE REPORT

J Korean Dent Sci. 2015;8(1):36-40
http://dx.doi.org/10.5856/JKDS.2015.8.1.36
ISSN 2005-4742

Pediculated Fibrous Dysplasia in Maxillary Sinus: A Case Report

Jung Ho Kim, Baek Soo Lee, Yong Dae Kwon, Byung Joon Choi, Jung Woo Lee, Hyun Woo Lee, Do Seop Kim, Joo Young Ohe

Department of Oral and Maxillofacial Surgery, School of Dentistry, Kyung Hee University, Seoul, Korea

Fibrous dysplasia is a bone disorder characterized by progressive replacement of normal bone by fibrous bone tissue. Common involving sites of fibrous dysplasia are the skeletal system including long bones, ribs, craniofacial bones and the pelvis. If maxilla were affected by fibrous dysplasia, antrum is almost always involved. And fibrous dyplasia in maxillary sinus were followed the shape of bone. In our case, the lesion involves antrum but, its shape was different from typical fibrous dysplasia pattern of maxillary sinus. Therefore we report a case of monostotic pediculated fibrous dysplasia in the maxillary sinus with a review of literature.

Key Words: Fibrous dysplasia; Maxillary sinus

Introduction

Fibrous dysplasia is a benign intramedullary fibrous-bone lesion invading the long bones, ribs, craniofacial bones and pelvis. Fibrous dysplasia accounts for 2.5% of benign tumor and 7.5% of benign bone tumor. Being typically asymptomatic during its pathological progression, fibrous dysplasia is observed by skeletal malformation, pathological fracture, etc.[1]. Among potential pathological causes suggested so far, fibrous dysplasia is deemed originated from abnormal osteoblastic differentiation due to gain on cAMP concentration affected by genetic mutation associated with G-protein[2]. Fibrous dysplasia is clinically categorized into monostotic fibrous dysplasia, polyostotic fibrous dysplasia and disseminated fibrous dysplasia, with monostotic fibrous dysplasia accounting for the greatest at the percentage of 80% to 85%. As for polyostotic fibrous dysplasia, McCune-Albright syndrome can be diagnosed when the bone lesion is accompanied by melanic lesion on skin or precocious pseudopuberty[3]. Oral and maxillofacial

Corresponding Author: **Joo Young Ohe**
Department of Oral and Maxillofacial Surgery, School of Dentistry, Kyung Hee University, 26 Kyungheedae-ro, Dongdaemun-gu, Seoul 130-701, Korea
TEL : +82-2-958-9440, FAX : +82-2-966-4572, E-mail : ojyoung81@hanmail.net

Received for publication February 11, 2015; Returned after revision June 6, 2015; Accepted for publication June 20, 2015

fibrous dysplasia is most commonly manifested in maxillary bone[4,5]. And fibrous dysplasia in maxilla commonly involves the antrum. Fibrous dysplasia in the maxillary sinus displaces the floor and follows its normal contour[6].

In this connection, this study is to discuss and bibliographically consider monostotic fibrous dysplasia pediculated in maxillary sinus.

Case Report

A 31-year-old patient, female, visited us with the chief complaint of the lesion of radiopacity in the left maxillary sinus, recognized upon panoramic examination conducted by a local clinic (Fig. 1A). Having no aesthetic and functional problems, the patient complained nasal stuffiness. Left maxillary teeth were viable with no paresthesia observed. No change in skin color or texture, traumatic history and hyperthyroidism were observed.

According to panoramic view, periapical radiography and Water's view, lesions with well-defined boundary in the left maxillary sinus and heterogeneously mixed radio-opacity were observed. No loss of lamina dura at molar teeth, external root resorption and displacement of teeth were observed (Fig. 1A). According to cone-beam computed tomography (CBCT), the lesions were linked to the left anterior maxillary sinus at zygomatic process of maxillary bone, with well-defined boundaries and pediculated lesions locally proliferated into the maxillary sinus. Partially mucosal thickening of maxillary sinus were observed as well (Fig. 1B). According to bone scan,

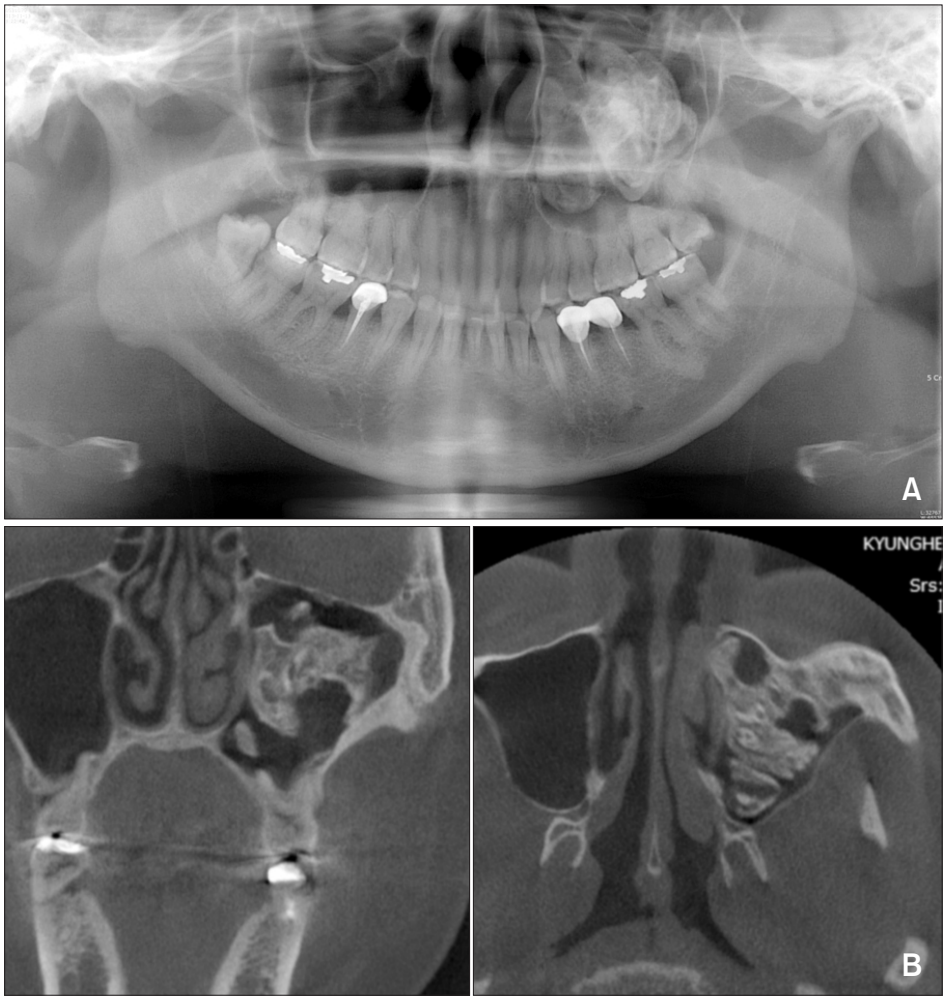

Fig. 1. (A) Radio-opaque lesion observed with well-defined boundary inside of left maxillary sinus. (B) Pediculated lesions locally proliferated into maxillary sinus with clear boundary against ground glass. No further invasion into any other structures was observed.

slight uptake increase at the left maxillary sinus were observed.

Upon general anesthesia, the patient received a surgical operation to remove lesion via maxillary anterior perforation. The lesion was observed to be tinged with gray-white with solidity (Fig. 2). No maxillary attachment was observed, save for anterior wall where is internal surface of zygomatic process. A sound ostium in maxillary sinus was observed after the surgical operation.

Histopathologically, the lesion featured a substrate of loose, cellular and fibrous tissue, with immature woven bone spread all across. Neither boundaries and capsules nor edges of osteoblast were observed, with the fibrous dysplasia featuring shape of a 'Chinese character' specific to it (Fig. 3).

Immediately after the surgical operation, the patient complained paresthesia in the left maxillary teeth, with decreasing scope of paresthetic regions and improved nasal stuffiness according to 3-month follow-up. According to radiographic examination, no lesional growth and abnormal change in maxillary sinus and mucosal thickening were observed (Fig. 4).

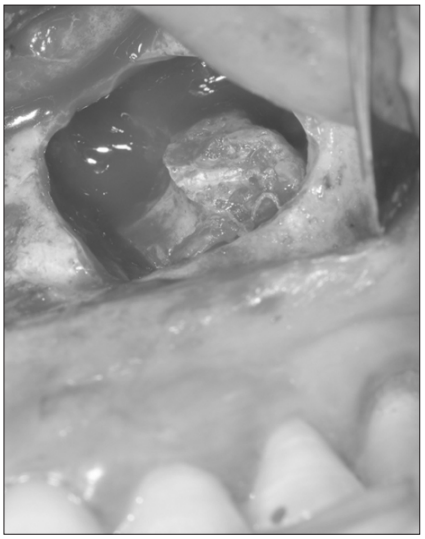

Fig. 2. Lesion with comparable solidity to gray-white cortical bone.

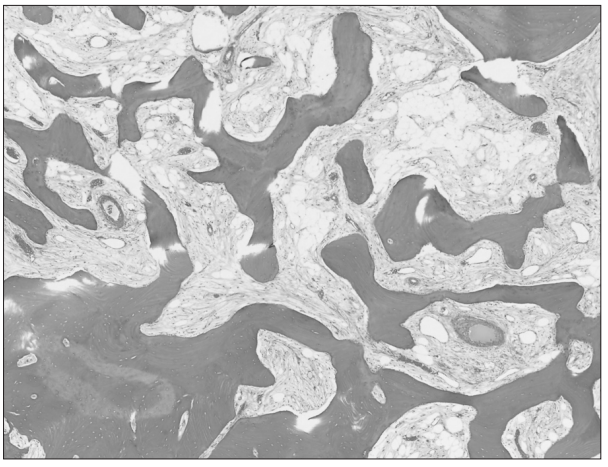

Fig. 3. Fibrous tissues and bone structures mingled to shape a 'Chinese character' specific to it (H&E staining, ×400).

Discussion

Discussed at the first time by Von Recklinghausen and granted a nomenclatural definition by Lichenstein back in 1938 , fibrous dysplasia had had equivocality in histological definition until 1942 when Lichenstein and Jaffe clinically classified fibrous dysplasia into monostotic fibrous dysplasia and polyostotic fibrous dysplasia, which were recently supplemented by disseminated fibrous dysplasia thanks to Feldman et al.[5]. A patient of disseminated fibrous dysplasia, referred to as McCune-Albright syndrome, suffers from symptoms irrelevant to bones such as sexual precociousness or café-au-lait skin macules and endocrinopathies[2]. Among the entirety of fibrous dysplasia patients, 25% of them are observed to have craniofacial lesions observed most commonly in jaw

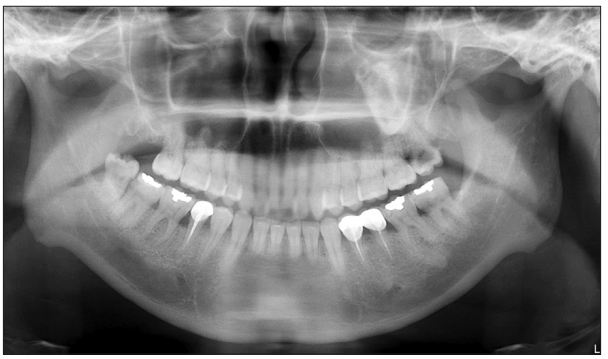

Fig. 4. Post-operative panoramic view.

bones, maxillary areas to be specific. Our case was observed to have no increase in uptake according to bone scan, to be safe for the maxillary bone. With no such irrelevant symptoms to bones that are specific to disseminated fibrous dysplasia, our case could be classified into monostotic fibrous dysplasia.

Fibrous dysplasia of maxilla has various and complicated appearance, reflecting the maxilla's complex anatomical structure. Antrum is frequently involved[6]. Although various external bone surface expansion pattern, internal expansion into antrum is almost typical[7]. Generally, fibrous dysplasia in maxillary sinus elevates its floor and reduces the volume of the antrum. Maxillary sinus could be completely or partially obliterated[6]. Further progress may lead to the displacement of orbital and nasal cavity[8]. This case showed a proliferative pattern towards maxillary sinus from zygomatic process of maxilla. It is the pedicle which connects the sinus lesion and the maxillary lesion. This pattern was considered different from typical fibrous dysplasia in the maxillary sinus.

Since it hardly accompanies pains, fibrous dysplasia is barely observed in the early stage. When metastasized to pelvis, the patient may easily suffer from fatigue fracture, pathological fracture and bone deformity where stresses are concentrated, which is how patients of young age around the age of 5 through 15 most commonly recognize fibrous dysplasia metastasized to pelvis. When it comes to facial bones, fibrous dysplasia happens to be recognized during clinical examination of facial swelling or from dental radiography. Specific type of pains resembling trigeminal neuralgia, ophthalmoptosis and/or other neurological symptoms may further manifest. When ethmoid sinus or frontal bone is invaded, a patient may further suffer from loss or weakening of olfactory sense, double vision and impaired optic canal and, when temporal bone is invaded, disability in auditory nerve and or vestibular nerve[2,9]. In the present case, lesion of maxillary sinus had been observed from the panoramic radiography for routine dental treatment, of which extension was further defined in infraorbital canal by way of CBCT. No neurological symptom, such as facial paresthesia, was observed.

With the maxillary sinus symptom improvement upon removal of lesion, it is deemed that difficulty in drainage was attributed to the symptom as ostium and the close proximity of the lesion with maxillary sinus. According to radiographic features, fibrous dysplasia can be classified into sclerotic fibrous dysplasia, cystic fibrous dysplasia and pagetoid fibrous dysplasia. Pagetoid fibrous dysplasia is the most common one as it features alternate radiolucency and radiopacity and full-scale bone expansion (56%), followed by sclerotic fibrous dysplasia (23%) featuring homogeneous opaqueness in ground glass and cystic fibrous dysplasia (21%) featuring well-defined sclerotic boundaries[10]. It is known that CBCT is useful in diagnosis and follow-up with accurate evaluation of detailed bone structures and lesional scope feasible. Despite tentative diagnosis as osteoma according to general radiography, our case was later diagnosed as fibrous dysplasia, classified as pagetoid fibrous dysplasia, according to CBCT for evaluation of external and internal bone structures. It is also known that the fibrous dysplasia seen as heterogeneous ground glass in 70 to 130 Hounsfield unit under CT enables the determination of invasion into bones[8]; however, the present case examined in CBCT did not show such invasion by way of Hounsfield unit.

From histopathological point of view, the lesion was observed to be tinged with gray-white with soft or solid intra-fibrous structure, and coarse and solid cross-section. From histological features, the lesion features shape of a 'Chinese character' which is specific to the fibrous dysplasia, with bony spur of irregular structures distributed across fibrous interstitium. Proliferating fusiform cell and bony spur without edges of osteoblast have been shown

to be specific to the interstitial structure[11]. Final diagnosis of fibrous dysplasia for our case was made based upon the specimen.

A single lesion needs no treatment if histologically diagnosed and should stand test of time until proliferation of lesion is halted for surgical treatment approach. If it accompanies functional and/or aesthetic problems such as exophthalmos, impaired vision or facial deformation, induced symptoms or evidences of malignancy, the lesion needs to be surgically removed. The necessity of surgical intervention and the scope of excision must be determined by taking lesional position, proximity to the nearest structures and symptomatic degree into consideration. Excision may take place by way of partial removal or excochleation. Administration of bisphosphonate formulation is worth consideration as well, when it comes to proliferating lesions[12]. Our case needed biopsy of tissues with proliferating appearance for decisive diagnosis and it was highly likely to be associated with the lesion proximate to maxillary sinus ostium, according to the symptomatic property of the chronic maxillary sinus. According to the improvement of the maxillary sinus symptom upon removal of lesion, it is deemed that difficulty in physical drainage in maxillary sinus ostium was attributed to the lesion.

Conflict of Interest

No potential conflict of interest relevant to this article was reported.

References

1. Edgerton MT, Persing JA, Jane JA. The surgical treatment of fibrous dysplasia. With emphasis on recent contributions from cranio-maxillo-facial surgery. Ann Surg. 1985; 202: 459-79.
2. Chapurlat RD, Orcel P. Fibrous dysplasia of bone and McCune-Albright syndrome. Best Pract Res Clin Rheumatol. 2008; 22: 55-69.
3. Kruse A, Pieles U, Riener MO, Zunker Ch, Bredell MG, Grätz KW. Craniomaxillofacial fibrous dysplasia: a 10-year database 1996-2006. Br J Oral Maxillofac Surg. 2009; 47: 302-5.
4. Pound E, Pickrell K, Huger W, Barnes W. Fibrous dysplasia (ossifying fibroma) of the maxilla: analysis of 14 cases. Ann Surg. 1965; 161: 406-10.
5. Feldman MD, Rao VM, Lowry LD, Kelly M. Fibrous dysplasia of the paranasal sinuses. Otolaryngol Head Neck Surg. 1986; 95: 222-5.
6. MacDonald DS. Maxillofacial fibro-osseous lesions. Clin Radiol. 2015; 70: 25-36.
7. Macdonald-Jankowski DS, Li TK. Fibrous dysplasia in a Hong Kong community: the clinical and radiological features and outcomes of treatment. Dentomaxillofac Radiol. 2009; 38: 63-72.
8. MacDonald-Jankowski DS. Fibro-osseous lesions of the face and jaws. Clin Radiol. 2004; 59: 11-25.
9. Chapurlat RD, Meunier PJ. Fibrous dysplasia of bone. Baillieres Best Pract Res Clin Rheumatol. 2000; 14: 385-98.
10. Hocaoglu E, Yirgin IK, Aksoy S, Arslan G, Inci E, Cimilli T. Computed tomography imaging findings of craniofacial fibrous dysplasia. J Craniofac Surg. 2014; 25: 1175-7.
11. DiCaprio MR, Enneking WF. Fibrous dysplasia. Pathophysiology, evaluation, and treatment. J Bone Joint Surg Am. 2005; 87: 1848-64.
12. Chapurlat R, Meunier PJ. The nonsurgical treatment of fibrous dysplasia. Rev Rhum Engl Ed. 1999; 66: 1-3.