Focal osteoporotic bone marrow defects associated with a cystic change of the maxilla: a possible histopathogenetic background of simple bone cyst

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Abstract: A case of focal osteoporotic bone marrow defects (FOBMDs) with a simple bone cyst-like change arising in the bilateral anterior maxilla of a 9-year-old Japanese boy is reported. Two FOBMD lesions were simultaneously found by chance as small oval-shaped unilocular radiolucencies symmetrically located between the canines and lateral incisors on a panorama radiograph during dental treatment of the patient's right maxillary canine, which erupted obliquely as the canine tooth roots were laterally displaced by the lesions. The lesions were surgically extirpated under a clinical diagnosis of developmental jaw cysts. However, they were shown at surgery not to be cysts but instead bone-marrow-like tissues, though the right lesion contained a cavity space within it. Histopathologically, they were fatty marrow without hematopoiesis, and irregularly-shaped bony trabeculae and blood clots were seen scattered throughout the marrow. They were diagnosed as FOBMD (left) or FOBMD with simple bone cyst (right), although they did not contain hematopoietic marrows. Thus, their fully fatty change with some blood pools suggests that they were in the initial stage of developing into simple bone cysts. Based on these histological observations, we propose a new hypothesis of FOBMD as one of the histopathogenetic precursors of simple bone cyst.

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Key words: cystic jaw lesion, focal osteoporotic bone marrow defect, histopathogenesis, jaw bones, simple bone cyst

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Introduction

Focal osteoporotic bone marrow defect (FOBMD) is a rare osteolytic lesion of the jawbone. It was first described by Standish and Shafer as a localized radiolucent lesion, which histologically consists of hematopoietic marrow and varying amounts of fatty marrow (1). It most often involves the posterior regions of the mandible, especially the edentulous areas, among middle-aged women (1-8). Although it has been speculated to be caused by tooth extraction from which an extraction socket does not follow

the normal regeneration process, some cases without traumatic histories are also documented in the literature (3). Also, as a systemic disease, FOBMD is known to be one of the signs of sickle cell anemia, in which FOBMD is considered to be caused by hyperplastic bone marrow resulting in red blood cell production (9-10). In such cases, bone defects frequently occur bilaterally in the molar region of the mandible (11). However, most of the cases seem to occur due to local factors, although they are not well elucidated. Because the radiographic appearances of FOBMD are varied radiolucencies, ranging from those with

Fig. 1. Dental radiograph of focal bone marrow defects of the maxilla arising in a 9-year-old boy. (a) right side, (b) left side. The lesions were well-defined radiolucencies between the roots of the lateral incisor and canine in bilateral sides. The left lesion was accompanied by some degree of opacity. Tooth roots of these teeth were divergently dislocated around the lesions.

well-defined borders to poorly marginated ones (12-13), the clinical differential diagnoses of FOBMD can include residual dental infection, osteomyelitis, eosinophilic granuloma, central neoplasms, or simple bone cyst (2-3, 14-15).

This report describes an unusual case with bilateral FOBMDs in the anterior maxilla without any systemic background, though their histological features were not always typical of those of FOBMD because they had no hematopoietic activities and one of them showed cavity formation. Based on our histopathological examinations, we discuss a possible histopathogenetic process towards simple bone cyst from FOBMD through fatty change and hemorrhage in the marrow space.

Case report

A 9-year-old Japanese boy was referred to the Pedodontic Department of Niigata University Hospital because of oblique eruption of the right canine tooth in his maxilla. The canine erupted from the labial side, while its preceding

Fig. 2. Intraoral view of focal bone marrow defect of the right side of the maxilla during surgery. When a piece of the cortical bone was removed, there was a cystic cavity in the cancellous bone.

deciduous canine was still present. The left canine had already erupted into normal position. There was no expansion of the labial or palatal cortical bones in the bilateral canine regions. The patient's general health and development were good with no particular familial or personal history for local or systemic diseases, including traumatic injuries to the maxilla. Hematological examinations did not show any abnormal findings. On dental radiographs, there were nearly symmetrical oval-shaped radiolucent areas with minimal trabeculation between the roots of the lateral incisors and canines on bilateral sides (Fig. 1). Due to the presence of those radiolucent areas, both of the tooth roots were widely dislocated, though there was no resorption of tooth roots and the lamina dura was intact. These teeth were vital and there was no evidence of periodontal disease around them.

Under a clinical diagnosis of developmental jaw cysts, extirpation of the two radiolucent foci was performed. When the labial mucoperiosteal flaps were raised, cortical bone surfaces were intact in the both lesions. A small piece of the bone cortex was removed from the right lesion, and there was a vertically-located ovoid-shaped cavity circumscribed by cancellous bone tissue (Fig. 2). The cavity was almost empty, retaining small pieces of bone marrow-like tissue containing hard tissues on the upper side. Curettage of those hard tissues induced bleeding. On the other hand, there was no obvious cavity in the left lesion; instead, it was filled with dark reddish-brown soft tissues, which were removed. The postoperative course was uneventful for five years after surgery, and the curettage zones became as radiopaque as those of the surrounding cancellous bone areas.

Pathological findings

The surgical specimens from the right lesion were composed of fragments of cortical bone and bone marrow-like tissue. Histologically, in the inner surface of the cortical bone fragment, there was a thin layer of loose connective tissue which seemed to be lining the empty space. The innermost part of the connective tissue was condensed, and this finding suggested the presence of a simple bone cyst wall (Fig. 3a). The bone marrow-like tissue removed from the upper part of the cavity was histologically fatty bone marrow, scattering many blood clots (Fig. 3b), in which there was no obvious hematopoietic activity; bone trabeculae were irregular-shaped, indicating active bone remodeling (Fig. 3c). Within the fatty bone marrow, there were occasionally small foci of fibrous granulation tissue (Fig. 3d). These findings indicated that the right lesion was basically consistent with simple bone cyst, but the focal exposure of fatty bone marrow into the cavity space with some organizing processes indicated that it was a kind of FOBMD in the process of developing into simple bone cyst. The specimen taken from the left side lesion was histologically a solid bone marrow tissue without hematopoiesis as was seen in the right lesion, but there was no cystic space (not shown). Although there were no hematopoietic bone marrow tissues, the left lesion could be diagnosed as FOBMD in which fatty changes were extensive.

Fig. 3. Histopathological findings of focal bone marrow defect of the right canine region of the maxilla. Hematoxylin and eosin stain. (a) A cortical bone fragment; (b-d) contents of the lesion, (a, d) $\times 50$, (b) $\times 20$, (c) $\times 25$. There was thin connective tissue in the inner surface of the cortical bone of the right lesion, suggesting the possible transition towards simple bone cyst (a). The lesion contained fatty bone marrows scattering blood clots (b). In the fatty marrow, bone trabeculae were irregularly-shaped (c), and there were focal granulation tissue changes (d).

Since both of the two symmetrical lesions were radiographically identical, they were considered to be the same FOBMD in different stages.

Discussion

The primary histopathology of FOBMD is documented as a hematopoietic bone marrow zone with fewer amounts of bone trabeculae and without fatty changes. Although its histopathogenesis remains unknown, three major processes have been hypothesized (1, 3, 16-17). The first one is that FOBMD is a remnant from early embryonic bone marrow which has not been converted to mature bone marrows. The second is that it is hyperplastic bone marrow, which is reactive to some local stimulus, such as trauma, in an abnormal healing process. The third is that it is part of a systemic condition causing an increased demand for red blood cells and resulting in marrow hyperplasia. Thus, FOBMD has been considered as a rather focal-excessive hematopoietic zone within the bone marrow. The present case, however, was not always consistent with conventional FOBMD in terms of its histopathological features.

In the present case, both lesions showed almost the same radiographic features with different surgical findings in terms of the presence of a cavity space, even though their overall background histology of fatty bone marrows was similar to each other. This histopathological evidence indicates that these two lesions may be different developmental stages of one pathologic condition. If the right lesion with a cystic cavity was in a more advanced stage than that of the left one, simple bone cyst might possibly arise in the background of FOBMD via its process of developing into the fatty marrow, because the cavity space found in the right lesion was circumscribed by a

connective tissue wall which was facing luminal contents. This feature is usually seen in simple bone cysts.

Shankland *et al.* proposed a new etiology, demonstrating that FOBMD might be the earliest detectable form of bone marrow edema secondary to a malfunction of blood flow within the marrow. They suggested that FOBMD was an intermediary step between an ischemic event with the subsequent development of bone marrow edema and the subsequent possible development of ischemic osteonecrosis (18), which might lead to simple bone cyst. Matsumura *et al.* pointed out that there were some variations in the radiographic and histological features of simple bone cyst, and that these differences might be simply due to time factors (19). Thus, taking all these matters into consideration, it might be possible to consider that both lesions were sequential variations of the same pathological condition.

The bilateral, symmetrical, and anterior maxilla manifestation of FOBMD is very rare, and the anterior maxilla location in a child is also unusual as simple bone cyst. Only a few FOBMD cases located in the anterior part of the maxilla have been documented in the literature (12). In addition, the divergent tooth roots neighboring the lesions and their bilateral location may indicate some congenital background. Gordy *et al.* reported a similar case of FOBMD which occurred between the roots of the maxillary right canine and lateral incisor in a middle-aged woman (17). Although its clinical diagnosis was simple bone cyst, only fatty bone marrow tissues were surgically removed from the lesion. Wilson *et al.* also reported a FOBMD with cyst-like cavity and fatty bone marrow tissues (13). These two cases seem to be very similar to our case.

Under normal conditions, the bone marrow of the jaw bones starts to be replaced with adipose tissues at five to seven years of age (20), and red or hematopoietic marrow

tissues, which are rich in active blood supply, are usually located only in the maxillary tuberosities and molar areas in adults (21). In the present patient, who was 9 years old, the bone marrow around the lesions must have already been fatty. Although fatty bone marrow is relatively poor in vascularity, it may be vulnerable to external stimuli, such as trauma, bacterial invasion, vascular congestion, and reduction of blood flow, all of which lead to ischemic and inflammatory changes (18). Therefore, the anterior areas of the maxillary bone may be a favorable location for pathological change.

From the present investigation, FOBMD was shown to be possibly one of the initial conditions towards sequential development of simple bone cyst. It is therefore advisable to follow up such lesions in children, although FOBMD has been considered to require no particular treatment (22).

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