Case Report

Stafne's bone defect: a case report and review of literatures

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Abstract: Stafne's bone defect (SBD) or Stafne's bone cavity (SBC) is an uncommon bony defect occurred especially only at the lingual cortex of mandible. Clinically, patients with SBD are usually asymptomatic. In most cases, the defect is observed accidentally via X-ray panorama during other dental treatments. Here we presented a female with SBD and we reviewed relevant literatures on SBD, summarized the clinical characteristic and radiographic features with our experiences.

Keywords: Stafne's bone defect (SBD); Stafne's bone cavity (SBC); case report

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Introduction

Stafne's bone defect (SBD) is an uncommon mandibular defect. It is usually accidentally found via radiograph during other dental treatments because patients often present no abnormal clinical symptom (1). Through panoramic radiography an ovoid or round radiolucency can be observed near the angle of mandible, usually located between the inferior alveolar nerve and the lower margin of mandible. The diagnosis of SBD is not too difficult with the help of CT and MRI, but differential diagnosis with other cyst-like lesions in mandible is needed, such as ameloblastoma, residual cyst, periapical cyst or lesions from salivary gland (2). We experienced a female patient with SBD located in the right mandible. This study aims to report and summarize clinical and radiographic features, histopathology and treatment after reviewing previously reported literatures relevant to SBD to facilitate diagnosis for clinicians in clinical practice.

Case presentation

A 48-year-old female was found via panorama to have an ovoid radiolucency in her right lower jaw. The patient was referred to our clinic to treat her decayed teeth but was discovered to have this lesion without presenting any pain and numbness in the mucosa and the lip. She was hospitalized and CT scanning as well as microscopic observation on specimen obtained from surgical intervention were performed. Features such as location, size, border and margin were carefully examined. X-ray image showed a well-defined ovoid radiolucency with high-density margin (*Figure 1A*). The lesion was located near the angle of the right jaw. In particular, the lesion was located below the periapical area of 47 and 48, whose roots were intact without any resorption. Right inferior alveolar nerve perforated the lesion. CT image revealed that the lack of continuity of lingual cortex of the right lower jaw could be found, leaving the buccal cortex extremely thin. Besides, soft tissue could be observed in the lesion (*Figure 1B*). Pathological results showed that clot and bone tissue were found in the specimen obtained from surgical intervention (*Figure 1C*). Considering all the collected evidence, there was a diagnostic indication of SBD. At the 6-month follow-up after discharge, the patient was asymptomatic and had a good recovery.

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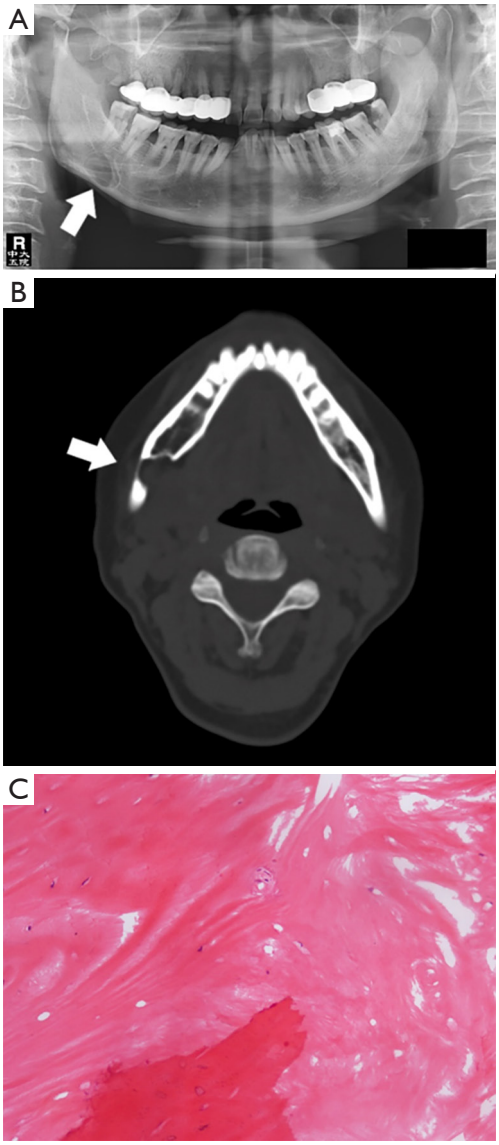

Figure 1 SBD in the right mandible. (A) Panorama image showed an ovoid radiolucency with hardened margin; (B) CT scanning further revealed lingual bony defect in the right mandible and soft tissue image was found in the defect; (C) pathological observation showed that clot and bone tissue were found in the specimen obtained from surgical intervention. Based on hematoxylin and eosin staining. Magnification: ×20. SBD, Stafne's bone defect.

Discussion

Epidemiology and origin

SBD is a rare lesion which was first reported in 1942 (3). The incidence is 0.1% to 0.48% in different reports (1). In fact, actual incidence of SBD may be higher than that's reported because of the difficulty of diagnosis without radiograph in cases where patients show no any abnormal symptoms. SBD are often observed among males between 50 and 70, while a reported youngest case occurred on an 11-year-old child (4). SBD can be observed in the posterior or anterior area of the mandible and the ramus of mandible which is very much rare. The etiology of SBD remains to be uncertain. It is mostly accepted that SBD is a developmental anatomic impression caused by proliferation or translocation of adjacent structures such as salivary gland or other soft tissues.

Clinical presentation

To summarize clinical and radiographic characteristic of SBD, relevant literatures search of PubMed for cases reports of SBD was carried out. Data such as gender, age, location of the lesion, manifestation, X-ray radiography and Computed tomography (CT) was retrieved and summarized in *Tables 1*,*2*.

In most cases, SBD is accidentally observed via panoramic X-ray film when patients are receiving other dental treatments. There are patients who come for help because they find an obvious lingual mucosal depression on their mandible. Pain is rarely found, so as to ulceration, bleeding or fistula. In our case presentation, patient was asymptomatic, which proved that SBD was uneasy to find without the help of radiographic examination.

Diagnostic imaging

Radiographic evaluation can greatly help to diagnose SBD. In our experience, panorama can provide initial evaluation of SBD. In most cases, a well-defined ovoid radiolucency can be observed beneath the inferior alveolar nerve. Apical area of canine and premolar is the most common location of SBD in the anterior area of mandible. Anterior SBD can be easily mistaken for residual cyst, which should raise enough attention in clinical practice. History of local tooth extraction after hopeless apical infection of relevant tooth can help to diagnose residual cyst while there is usually no contact of SBD with relevant teeth. CT has the ability to show more details when it comes to the buccolingual location and provide information on whether or not the defect has perforated through buccal cortex, which 2-dimentional panorama cannot provide. Further CT scanning after speculated diagnosis of SBD from panorama to determine buccolingual position does great help to the differential diagnosis of SBD (9,11). Three-

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Table 1 Individual case reports of literature review

Author	Patient's gender	Age	Location	Manifestation	X-Ray radiography	CT radiography
Lee *et al.* (5)	Female	52	Right angle of mandible	No symptom	Not mentioned	Cortical defect containing a soft tissue lesion
Venkatesh *et al.* (6)	Male [2]	31, 35	Left side of mandible	No symptom	Both are cystic lesions	Radiolucent lesions, regular, oval in shape, and well corticated.
Lee *et al.* (7)	Male	30	Right angle and ramus of mandible	Painless swelling of right face	Radiolucency on right ramus of mandible	A depression and lack of cortical surface
Taysi *et al.* (8)	Male	56	Apical area of 33-35	No symptom	Well-defined ovoid radiolucency	Lingual bone defect with thinning buccal cortex
Probst *et al.* (9)	Male	72	Left posterior area of mandible	No symptom	Well-defined ovoid radiolucency with hardened margin	Lingual bone pit
Kim *et al.* (10)	Female	44	Anterior area of mandible	No symptom	Two well-defined ovoid radiolucencies	Not mentioned
Saglam *et al.* (11)	Male	56	right angle of mandible	No symptom	Well-defined radiolucency	Lingual bone defect and thin buccal cortex with slightly lobulated expansion
Boffano *et al.* (12)	Male	72	Left body of mandible	No symptom	Radiolucency with irregular shape	Lingual trilobite bone defect
Munevveroglu *et al.* (4)	Male [2]	31, 57	Left angle of mandible	No symptom	Cystic radiolucency	Ovoid lingual bone defect
Etoz *et al.* (13)	Male	58	Left molar area of mandible	No symptom	Multilocular radiolucency	Irregular lingual bone depression
Dereci *et al.* (14)	Male	46	Left premolar area of mandible	Obvious lingual mucosal depression on left mandible (premolar area)	Not mentioned	Lingual bone depression
Li *et al.* (2)	Male	40	Below the 37	No symptom	Irregular radiolucency	Lingual bone defect
Sisman *et al.* (15)	Female	62	Right premolar area of mandible	No symptom	Well-defined unilocular radiolucency	Lingual bone defect
Kopp *et al.* (16)	Male	67	Apical area of 37	No symptom	Periapical radiolucency of 37	Not mentioned

Table 2 Group case report of literature review

Author	Sample size (patients)	Gender	Mean age	Location	Manifestation	X-ray	CT image
Sisman *et al.* (1)	29	Male =25, Female =4	49.6	Left =13, right =16	Not mentioned	Cystic radiolucency in mandibles	Unilateral lingual bone defect in molar-angle area with or without reaching buccal cortex

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dimensional reconstruction of CT is now available to help to directly observe the spatial relationship of SBD and the inferior alveolar nerve (2,7). However, though much benefits can be brought from CT radiograph, consequent ionizing radiation is still a worrying issue in CT evaluation. MRI can not only rid patients of ionizing radiation but also help to observe the content of the defect, which is beneficial to the diagnosis of SBD. The disadvantages of MRI may be relatively high cost, discomfort feeling during the scanning and the radiographic artifacts and distortion in images. Sialography is an effective technique to determine whether there is glandular tissue in the defect, which is an important indication of the glandular sources of the lesion. But it is unsuitable for anterior SBD because submandibular glandular ducts are so tiny that imaging in the ducts is almost impossible (5,8).

Differential diagnosis

SBD should be diagnosed excluding other odontogenic or non-odontogenic cystic lesion. Dentigerous cyst can also be observed near the angle of mandible, containing the crown of an un-erupted tooth mostly occurred in the impacted wisdom tooth. Radicular cyst is obviously related to inflamed teeth which could cause relevant symptoms on teeth such as bite pain or swelling. Ameloblastoma can present near the angle of mandible. Consequent cut-shaped root resorption of teeth can provide important clue to differentiate these two diseases as SBD have no relationship to the teeth. Langerhans cell histiocyosis (LCH) can be observed below the lower molars, whose radiolucency is usually ill-circumscribed. LCH is mostly found in children in 5-15 and could cause swelling and pain on maxillofacial area. Differential diagnosis also includes other jaw lesions such as vascular malformation, non-ossifying fibroma and fibrous dysplasia, basal cell nevus syndrome and a metastasis from a primary malignant tumour from other organs.

Management

As SBD is a benign, developmental bony defect causing no any pathological changes, surgical intervention is no longer needed to treat SBD. But follow-ups on a regular basis are recommended to check whether there is radiographic enlarging tendency or any abnormal signs of the lesion.

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None.

Footnotes

Conflicts of Interests: The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. Written informed consent was obtained from the patient for publication of this manuscript and any accompanying images.

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