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Case Report

Anterior stafne defect of the mandible

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Abstract
Stafne bone cavity is round or ovoid, well circumscribed radiolucency located at the lingual cortex of mandible, below the inferior alveolar canal, mostly near the angle. Anterior location of Stafne bone cavity is a unusual developmental anomaly and only 66 cases reported in the English literature. As a result of the localization of this formation, which is asymptomatic and diagnoses with routine radiographic examination, it may be confused with pathologies and may cause misdiagnosis. In this case report a Stafne bone cavity located in the mandibular incisor-canine region, which was found incidentally during radiographic examination cavity is presented with cone beam computed tomography and magnetic resonance images.

Keywords: Cone beam computed tomography; magnetic resonance imaging; mandible

INTRODUCTION

Stafne bone cavity (SBC) was first described by Stafne in 1942 as a round or oval shaped, well circumscribed, located lingually on the posterior mandible below the mandibular canal, asymptomatic lesion detected randomly in intraoral radiographs from patients (1). The other names for this lesion used in the literature are static/latent/idiopathic defect/cavity/cyst, developmental lingual mandibular salivary gland defect, developmental salivary gland defect, mandibular lingual cortical defect, aberrant/ectopic salivary gland, lingual mandibular bone cavity/concavity/defect/depressions, mandibular salivary gland inclusion, Stafne's cyst/defect/cavity (2,3).

This lesion had similar radiographic features since the first day of diagnosis and the lesion was found to have a wide variety of contents when surgically removed. It was found that this lesion consists of normal or inflamed submandibular or ectopic salivary gland tissue, normal or inflamed lymph gland tissue, fatty tissue, fibrous connective tissue, blood vessels, peripheral nerves or entirely void (4). Since SBC is not surrounded by a true epithelium, it is classified in the group of pseudocysts. And they have an incidentally detected cystic appearance in panoramic x-rays (4,5). Also some authors offer the term cavity instead of cyst due to the fact that this term is more accurate for their opinion (6).

In 1957 Richard and Ziskind have reported an anterior variant of SBC for the first time. They observed lingual defect in the premolar region of the mandible and called aberrant salivary gland tissue in the mandible (7). Then in 1980 Bucher et al. diagnosed anterior defects area in cuspid and incisor region of the mandible and they chose to name it the anterior lingual mandibular salivary gland defect (2). This lesion also usually involves ectopic salivary gland tissue and do not need surgery or biopsy (2,5).

From 1957 up to now only 66 cases of the anterior location of SBC were reported (3). For this reason; the aim of this report is to add a new case of SBC in the anterior mandible with clinical and radiographic features

CASE REPORT

A systemically healthy 45-year-old white man was admitted to Recep Tayyip Erdoğan University, Faculty of Dentistry, Oral and Maxillofacial Radiology Department for a pre-diagnosed cystic lesion located in the beneath the apices of lateral incisor and canine of the left mandible. A well-defined, unilocular lesion which is not related to the teeth roots was seen at panoramic radiography (Figure 1). The patient was informed that radiographic images could be used for scientific purposes and informed consent

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was obtained. Intraoral and extraoral examination showed no expansion, crepitation or fluctuation. The lesion was asymptomatic, and the patient was unaware of its presence. The patient's medical history was not significant.

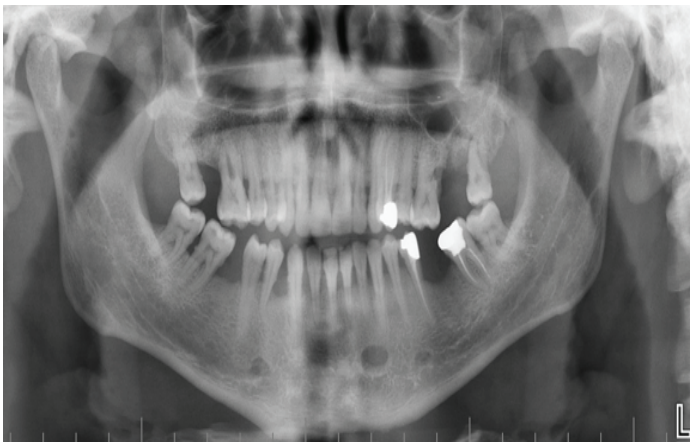

Figure 1. Ortopantomography shows a radiolucent corticated lesion to the left mandibular incisor- canine region

A cone beam computed tomography (CBCT) scan was obtained from the patient, a corticated, well defined, concave area below the root of the lateral incisor at the lingual area. The concave area reaches to the trabecular area, not to the buccal cortical plate, the defect classified as Type 1 SBC according to the Ariji et al. (Figure 2). The initial diagnosis was made as anterior SBC and patient called for an magnetic resonance imaging (MRI) examination to assess the area and confirm the diagnosis (8).

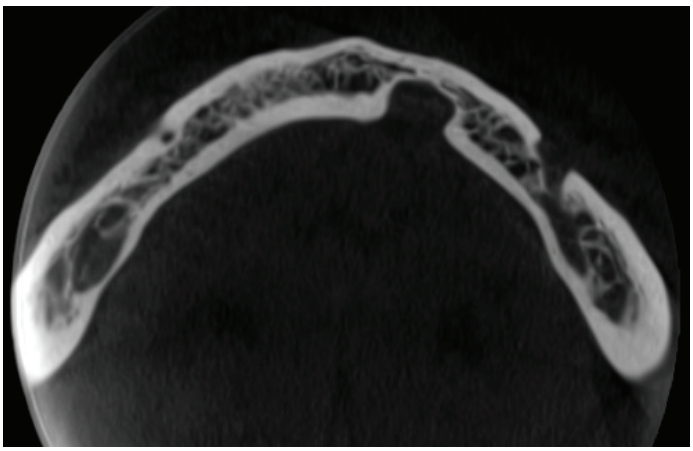

Figure 2. CBCT scan, axial view, shows the radiolucent area to be a corticate defect on the lingual surface of the mandible

T1- weighted and fat-saturated contrast-enhanced T1-weighted MRI scans showed the concave area located at the lingual side of the mandible filled with salivary gland tissue which is considered as a hyperplastic part of the sublingual gland (Figure 3). CBCT and MRI findings were compatible with an anterior variant of SBC. The lesion was diagnosed as anterior SBC and patient was called for a routine follow-up.

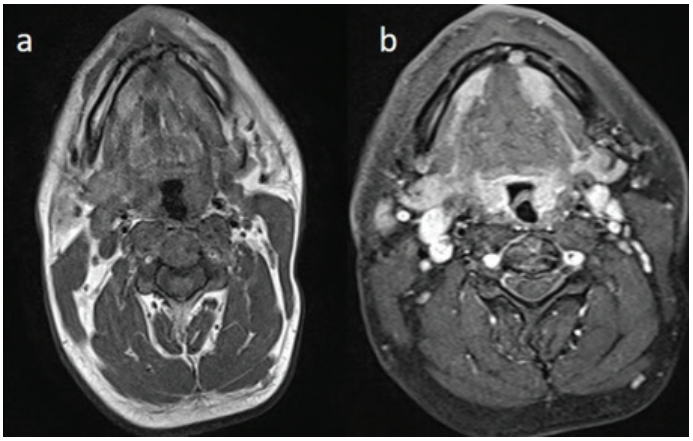

Figure 3. (a)T1-weighted MR image, axial view, shows that the mandibular defect contains soft tissue in continuity with, and isointense with, the sublingual salivary gland (b) On fat-saturated contrast-enhanced T1-weighted MRI, axial image, shows that the bony defect contains an extension of tissue from the sublingual salivary gland

DISCUSSION

The typical radiographic feature of SBC is a well-demarcated, oval or round-shaped uniform radiolucent appearance of the mandible under the mandibular canal (1). Rarely the lesion may show multilocular appearance and the borders may not always show clearly defined (9). The image of SBC may differ, Ariji et al. divided SBC's into three types according to the outline and relation with buccal cortical plate where in type I lesions concavity reach to the trabecular area, in type II the concavity reaches to buccal cortical plate and in type III concavity reach buccal cortical plate and shows an expansion (8). The lesion may superimpose to teeth roots or may locate beneath the apices, rarely between roots and edentulous areas. The defect size was reported between 0,5 mm to 2 cm (3). SBC was seen in more frequently in males with a ratio between 6:1 and 3:1 (10). The decade for highest incidence for genders differs whereas in males the highest incidence was seen fifth to sixth decades, in females between third and fifth decades for SBC's. In male patients reported cases are between 18-68 age with the mean age of 43 (3). In literature no patient was reported with Stafne bone cavity younger than 11 years old (10). Anterior SBC's prevalence is estimated at 0.009%-0.3%. The male/female ratio was reported as 3/1 for anterior SBC's (9). Our patient's age is in accordance with the literature and also very close to the mean value.

66 anterior SBC reports have been detected until today, the lesion is usually found in the canine-premolar region (3). It was observed that 4 cases were found in the lateral incisor and canine region (10,11). In reported cases located in the lateral-canine area, only Bornstein et al.'s case has tomographic images and like our one, the cavity reaches to the trabecular area of the mandible and not to the buccal cortical plate (10). Our case seems that it is the fifth anterior SBC seen in the lateral-canin region in the literature.

SBC may be seen at different locations as above the mylohyoid muscle at the lingual surface of incisor/canine/premolar, posterior to the mandibular angle-first molar area, below mandibular canal, buccal of ascending ramus, at ascending ramus posterior of the mandibular foramen. SBC's mostly unilateral but bilateral cases were also rarely reported. Most of the reported anterior cases were located between the cuspid and the first molar (57%). Fewer cases involved the incisor area (26%). The remaining (17%) concerned more extensive lesions spreading through the anterior mandible (3). SBC has the anterior and posterior variant. The posterior variant is seen between mandibular premolar region and angulus mandible, anterior variant is generally located between cuspid and first molar teeth, above the mylohyoid muscle and beneath the root apices if the teeth are present (12,13). Similar to posterior SBC, anterior SBC is asymptomatic, non-progressive and rarely the lingual defect can be clinically palpated (14). In contrast to defects in the posterior region, defects in the anterior region are more difficult to diagnose. Because the bone defect in the anterior region can be confused with cystic lesions (such as a radicular cyst, residual cyst), that may be present in this region (13). Also traumatic bone cyst, lateral periodontal cyst, early stage focal cemento-osseous dysplasia must come to mind for a differential diagnosis. The misdiagnosis leads to incorrect treatment options like endodontic therapy, bone exploration surgery or bone trephination (10). In addition to these, archaeological studies showed bilateral depressions at the anterior buccal area of the mandible named as anterior buccal mandibular depression which is most prominent at pediatric populations that may be mistaken for anterior SBC's (4).

Pathogenesis of the lesion is somewhat controversial where some authors believe this defect is congenital and some authors believe it has a developmental nature (3). Due to no reported cases younger than 11 years old present the congenital theory is questionable (10). Constant pressures from neighboring structures like a submandibular gland or facial artery were suggested as a determinant factor. Also entrapment of salivary gland tissue during mandibular development was suggested for the formation of the defect (3). The most widely accepted theory for the posterior variant is presence of hyperplastic/hypertrophic lobe of submandibular gland whereas for anterior variant the widely accepted theory is the presence of aberrant lobe of sublingual gland (10). Embryonic rests of salivary glands may be present in jawbones which was shown in literature and this may be the etiological factor for Stafne defects that have an intact lingual cortex (6).

The diagnosis of this lesion is usually made by taking routine panoramic radiographs. CBCT, MRI and sialography radiography techniques are more important in the differential diagnosis of the anterior SBC (6,8,15). Causes of the difficulty in cannulation of the ducts of Rivini, high failure rate, possible sepsis, acinar rupture and duct trauma in the diagnosis of anterior SBC limits the use of sialography (3,6). CBCT scan is easy to perform and noninvasive, capable of demonstrating size, extent and 3-dimensional information of lesion. Furthermore, it is an important method used in diagnosis because the cortical boundaries in the lingual region of the mandible with CBCT can be clearly seen but ionizing radiation exposure was a significant risk and it does not allow examining soft tissues in detail. In addition to the soft tissue imaging and non-invasive method, MRI can be used to identify the lesion with the advantage that the patient is not exposed to contrast material and ionizing radiation. The disadvantages of MR imaging are the high cost and metal and distraction artefacts, as well as claustrophobia in the patient (15). In MRI the sublingual salivary gland is often continuous to the lesion and of equal signal intensity to the tissues adjacent to the bony defect (3). Also ultrasound imaging may be an option for assessment of the areas but as the high technical skill required for both scanning and interpreting of the relevant area, significant errors may be seen; therefore, this limits the clinical usage (4).

Most of the SBC lesions are asymptomatic and non-progressive (9). No treatment proposed for the area but taking a biopsy may be necessary where the area shows symptoms or an additional pathology suspected at the relevant area (3). In literature only 1 case reported that pleomorphic adenoma was presence in the posterior variant of Stafne defect (4). We think that MRI scanning which has no known harmful effect is necessary not for just atypical cases, to rule out possible pathology that can be located inside the cavity.

In our study, the lesion was asymptomatic and it was an incidental finding on routine clinical examination. CBCT and MRI were observed in accordance with SBC radiographic features. Therefore surgical excision is not considered. Correspondingly, the diagnosis was made as Stafne bone cavity and patient was called for a routine follow-up.

CONCLUSION

In conclusion; the SBC should be kept in mind when diagnosing radiolucent lesions that are usually clinically asymptomatic, which are rarely seen in the anterior part of the mandible compared to the posterior part of the mandible. Only panoramic radiography is not sufficient for the diagnosis of the anterior variant of SBC; the diagnosis should be supported as one of the advanced imaging methods. Clinical and radiographic features of anterior SBC should be known to prevent misdiagnosis and confusing other pathological lesions. No treatment is necessary for asymptomatic cases of SBC, radiographic follow-up is sufficient.

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REFERENCES

1. Stafne EC. Bone cavities situated near the angle of the mandible. J Am Dent Assoc 1942;29:1969-72.
2. Buchner A, Carpenter WM, Merrell PW, Leider AS. Anterior lingual mandibular salivary gland defect: evaluation of twenty-four cases. Oral Surg Oral Med Oral Pathol 1991;71:131-6.
3. Orhan K, Rozylo-Kalinowska IK, Yetimoglu-Ozdil N, et al. Stafne bone cavity in the anterior mandible: report of rare cases and literature review. Chirurgia Italy. 2018;31:90-8.
4. Smith MH, Brooks SL, Eldevik OP, et al. Anterior mandibular lingual salivary gland defect: a report of a case diagnosed with cone-beam computed tomography and magnetic resonance imaging. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2007;103:71-8.
5. Regezi JA, Sciubba JJ, Jordan RC. Oral pathology: clinical pathologic correlations. 6th ed. St. Louis, Missouri: Elsevier Health Sciences; 2012.
6. de Courten A, Küffer R, Samson J, Lombardi T. Anterior lingual mandibular salivary gland defect (Stafne defect) presenting as a residual cyst. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2002;94:460-4.
7. Richard EL, Ziskind J. Aberrant salivary gland tissue in mandible. Oral Surg Oral Med Oral Pathol. 1957;10:1086-90.
8. Ariji E, Fujiwara N, Tabata O, Nakayama E, Kanda S, Shiratsuchi Y, et al. Stafne's bone cavity. Classification based on outline and content determined by computed tomography. Oral Surg Oral Med Oral Pathol 1993;76:375-80.
9. Taysi M, Ozden C, Cankaya B, et al. Stafne bone defect in the anterior mandible. Dentomaxillofac Radiol 2014;43:20140075.
10. Bornstein MM, Wiest R, Balsiger R, et al. Anterior Stafne's bone cavity mimicking a periapical lesion of endodontic origin: report of two cases. J Endod 2009;35:1598-602.
11. Anneroth G, Berglund G, Kahnberg KE. Intraosseous salivary gland tissue of the mandible mimicking a periapical lesion. International journal of oral and maxillofacial surgery. 1990;19:74-5.
12. Philipsen HP, Takata T, Reichart PA, Sato S, Suei Y. Lingual and buccal mandibular bone depressions: a review based on 583 cases from a world-wide literature survey, including 69 new cases from Japan. Dentomaxillofac Radiol. 2002;31:281-90.
13. Katz J, Chaushu G, Rotstein I. Stafne's bone cavity in the anterior mandible: a possible diagnostic challenge. Journal of endodontics. 2001;27:304-7.
14. Dereci Ö, Duran S. Intraorally exposed anterior Stafne bone defect: a case report. Oral Surg Oral Med Oral Pathol Oral Radiol 2012;113:1-3.
15. Branstetter BF, Weissman JL, Kaplan SB. Imaging of a Stafne bone cavity: what MR adds and why a new name is needed. AJNR Am J Neuroradiol 1999;20:587-9.