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Fifteen-millimeter Stafne defect in the anterior mandible: Diagnostic contribution of multislice computed tomography

Abstract:

A Stafne bone defect is a cavity in the mandible. There is controversy regarding the cause of this defect, but imaging findings offer important diagnostic contributions. The aim of this report is to present the tomographic findings of a rare case of an extensive bone defect in the anterior mandible. A 55-year-old man was seen at a dental service for oral rehabilitation with dental implants. He was asymptomatic. Panoramic radiography revealed an oval, delimited radiolucency of cystic and regular appearance encompassing the area from the apex of tooth 26 to tooth 29; the major axis was parallel to the base of the mandible. Multislice computed tomography showed a cavity with heterogeneous content and the following measurements: buccal-lingual width/depth, 1.0 cm; anteroposterior width, 1.38 cm; height, 1.54 cm; area, 0.94 cm²; volume, 2.14 cm3; and attenuation coefficient \pm standard deviation, 19.86 \pm 91.27 HU. These data confirmed the diagnosis of an anterior variant of Stafne bone defect. The size of a Stafne bone defect needs to be considered when planning implant-supported rehabilitation and other interventions in the affected area. Recognition and definitive diagnosis of this condition are essential, particularly with increasing use of rehabilitation treatments with osseointegrated implants.

Keywords: X-Ray Computed Tomography; Radiography, Panoramic; Salivary Glands; Diagnostic Imaging

INTRODUCTION

Stafne bone defect, Stafne bone cavity, or Stafne cyst was first described by Stafne in 1942 as a bone cavity in 34 patients; most of these defects were unilateral and located in areas near the angle of the mandible. The cavities were discovered incidentally by the author on intraoral radiographs in the absence of symptoms or other relevant clinical findings¹. Radiographically, these cavities appear as well-delimited circular or oval radiolucencies measuring 1–3 cm in diameter¹.

Because of their asymptomatic nature, Stafne bone defects are diagnosed during routine examinations and have been included in the study of incidental findings. These defects are uncommon compared with other findings such as mucosal thickening of the maxillary sinus, the presence of impacted teeth, and calcification of the stylohyoid ligament, even in studies involving a considerable sample^{2,3}. Some theories on the origin of this defect have been suggested, including failure of ossification of Meckel's cartilage¹, pressure exerted by hypertrophy/hyperplasia of the salivary glands⁴, and a vascular neoplasm or lipoma⁵.

Stafne bone defects are generally found in the posterior mandible, inferior to the mandibular canal, but may occur at other sites such as the mandibular ramus⁶ or anterior mandible⁷⁻¹⁰. The radiographic features of these defects sometimes resemble true cysts^{11,12}. The diagnosis of a Stafne bone is challenging when it is located in the anterior mandible because occurrence is rare at this site, where only 63 cases of Stafne bone defect have been reported so far¹⁰. In addition, the similarity of the radiographic features to other equally radiolucent and well-delimited lesions, such as inflammatory radicular lesions, cysts, or benign tumors¹³, may lead to inadequate therapeutic management.

Two-dimensional radiographic images have been shown to be efficient in cases of classic posterior Stafne bone defects. However, in doubtful cases or cases in an unusual location, more accurate imaging methods such as computed tomography (CT) and magnetic resonance imaging are required to provide a correct diagnosis and to permit imaging follow-up^{14,15}. Therefore, the aim of this study was to discuss the tomographic features of a rare case of an extensive Stafne bone defect in the anterior mandible.

CASE REPORT

A 55-year-old man was seen at a private clinic for oral rehabilitation with dental implants. No pathologies or significant alterations were identified on physical examination and he was asymptomatic. For the initial assessment, a panoramic radiograph was obtained that demonstrated an oval, well-delimited radiolucency of cystic and regular appearance encompassing the underlying area from the apex of tooth 26 to tooth 29 with the major axis parallel to the base of the mandible (Fig. 1). Multislice CT was performed for further investigation. Multiplanar reconstruction images, attenuation coefficients (mean Hounsfield units $[HU] \pm$ standard deviation), volume, area, anteroposterior width, height, and buccolingual width/depth of the lesion (Table 1; Figs. 2 and 3) were obtained using Osirix DICOM Viewer software (Pixmeo SARL, Bernex, Switzerland).

Based on these reconstructions, a bone defect with lingual continuity and regular and corticated margins occupying almost the whole right mental region was identified. The defect measured approximately 1.5 cm at its greatest extension (height), with a thin rim of buccal cortical bone and low but variable density. The defect was classified as type I-S according to the classification proposed by Ariji et al.¹⁶ (Table 2).

DISCUSSION

The term "lingual and/or buccal bone depression" as suggested by Philipsen et al.⁴ is appropriate to encompass all variants of this defect, considering that its occurrence in the anterior region, as reported here, was not described in the first case series published in the literature. However, various names have been given to this entity, such as Stafne bone cavity¹³, Stafne defect¹⁷, and Stafne bone cyst¹⁵.

Most of the first cases reported were confined to the medullary portion of bone. However, defects with large diameter extended to the external cortical bone and interrupted the continuity of the inferior border of the mandible¹. In the present case, the bone defect was in contact with the buccal and inferior cortical bones but their integrity was preserved. In addition, measurement of the attenuation coefficient allowed the content of the cavity to be estimated; it was compatible with fat tissue

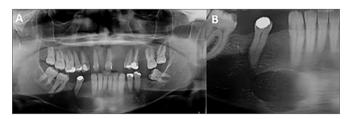


Figure 1. Panoramic radiograph. (A) Unilocular radiolucency beneath the roots of teeth 26 to 29. (B) Close-up view of the right mental region.

Table 1. Measurements of the bone defect obtained by multiplanar
reconstruction of the tomographic scan.

	Mean
Buccolingual width/depth (cm)*	1.00
Anteroposterior width (cm)	1.38
Height (cm)	1.54
Area (cm ²)*	0.94
Volume (cm3)	2.14
Mean density \pm standard deviations (HU)*	19.86±91.27

*Value obtained in axial section.



Figure 2. Multislice computed tomography. (A) Axial section showing a lingual bone defect with a slight reduction of the buccal cortical plate and defined corticated margins. Note the value of 19.866 HU for the content compared with the fat areas (–8 HU) adjacent to the lingual cortical plate of the mandible in the posterior region. (B) Coronal section. Observe the extensive bone defect confined to the medullary portion of bone, preserving the base of the mandible.



Figure 3. Multislice computed tomography. (A) Segmented volume. Reconstruction of the cavity volume highlighted in blue. (B) Three-dimensional reconstruction. Lingual view. The red arrows indicate the circular bone defect located in the right anterior mandible. (C) Reconstruction with soft tissues. Note the image is suggestive of fat and muscle tissues invading the bone cavity.

possibly intermingled with glandular tissue, muscles, vessels, and arteries of the region, as indicated by the high standard deviation recorded (± 91 HU).

According to the classification proposed by Ariji et al.¹⁶, the defect observed here corresponds to type I-S (Table 2). This important, simple, and straightforward classification has been used in different studies^{11,17-19}. To encompass all variants of the defect, we recommend the modifications shown in Table 2.

In 2016, a retrospective analysis of 999 conebeam CT scans regarding incidental findings, such as carotid artery calcification, paranasal sinus findings, sialolithiasis of the parotid and submandibular glands, calcification of the stylohyoid ligament, and Stafne bone

Table 2. Modification	of the classificat	ion proposed b	y Ariji et al. ¹⁶ .

According to the Outline and Relationship to the Buccal Cortical PlateTypeAccording to the ContentICavity depth is limited to the medullary portion of the mandibleFCavity is filled with fatIICavity depth reaches the buccal cortex of the mandible but does not cause its expansionFCavity is filled with softIIICavity depth reaches the buccal cortex of the mandible and causes its expansionSCavity is filled with softIIICavity depth reaches the buccal cortex of the mandible and causes its expansionGCavity is filled with some glandular tissue (not exclusively the submandibular gland)					
I the medullary portion of the mandible F Cavity is filled with fat II Cavity depth reaches the buccal cortex of the mandible but does not cause its expansion S Cavity is filled with soft tissue (lymph node, vessel, connective tissue, etc.) III Cavity depth reaches the buccal cortex of the mandible and causes its S Cavity is filled with soft tissue (lymph node, vessel, connective tissue, etc.)	Туре	Relationship to the Buccal	Туре	According to the Content	
IIthe buccal cortex of the mandible but does not cause its expansionCavity is filled with soft tissue (lymph node, vessel, connective tissue, etc.)IIICavity depth reaches the buccal cortex of the mandible and causes itsCavity is filled with soft tissue (lymph node, vessel, connective tissue, etc.)	I	the medullary portion of the	F	Cavity is filled with fat	
III the buccal cortex of the mandible and causes its G (not exclusively the	11	the buccal cortex of the mandible but does not cause	S	tissue (lymph node, vessel,	
	111	the buccal cortex of the mandible and causes its	G	some glandular tissue (not exclusively the	

defect, found 350 incidental findings and no case of Stafne bone defect³, demonstrating that its rare occurrence in the general population is rare, with an average prevalence of 0.08% in the posterior region and 0.003% in the anterior region¹⁴.

A previous study evaluating 34,221 panoramic radiographs reported a higher prevalence of Stafne bone defects in individuals older than 40 years and in men, with unilateral presentation and a slight predilection for the right side of the mandible¹⁴. In that study, only one of the 29 cases identified was located in the anterior region (near the premolars).

In the latest study on this defect²⁰, among the 91 cases of Stafne bone defect that were classified into three variants, only 2% were classified as the anterior variant, with the following characteristics: unilocular, partial radiolucency, oval shape, partially or completely sclerotic margins, and localization between premolars. Both studies reinforce the rarity of the case reported here in terms of localization and size, with a mean height and width of the cavities in the anterior region of 7.05 and 11.6 mm, respectively. In the present case, a height of 15.4 mm and width of 13.8 mm were observed.

There is controversy regarding the cause of this defect. Stafne¹ suggested failure in normal bone deposition during the neonatal period as the cause of the defect. Later, an extensive literature review suggested Stafne bone defects were the result of the pressure exerted by hypertrophic/hyperplastic glandular tissue on the bone surfaces⁴.

This theory was refuted by the reported occurrence of the defect on the lingual surface of the mandibular angle where the medial pterygoid muscle is inserted, preventing contact between the submandibular gland and bone⁶. At the most common site of the defect, the associated salivary gland is the submandibular gland. In the present case involving the anterior mandibular region, the bone defect could be associated with the sublingual gland.

However, because its volume is much lower than that of the submandibular gland, there are doubts about the possibility of the sublingual gland exerting enough pressure to cause such an extensive defect. In addition, the large standard deviation observed for the internal density of the defect and its anatomic position suggest the presence of muscle (35–60 HU) in the lesion. The analysis is the images suggest that the main muscle involved is the genioglossus muscle, based on the volume and location with central participation of the genioglossus. In view of these considerations, cone-beam CT can provide limited information for the diagnosis of this defect and doubts about the diagnosis would possibly remain.

In the absence of a relationship between the cavity and important anatomic structures, for example, the mandibular canal, there is a strong suggestion of characteristics similar to common bone pathologies such as inflammatory cysts. In edentulous areas as in the case reported here, Stafne bone defects can easily mimic residual cysts¹² or other pathologies. However, characteristics such as the presence of sclerotic margins and the absence of cortical bone expansion and tooth displacement, data provided by multislice CT, contribute significantly to the diagnosis of this bone defect. Other benign lesions with unilocular presentation, such as odontogenic keratocysts, may also be a suspected diagnosis. Once a Stafne bone defect is identified, twodimensional examinations should be repeated every 12 months to evaluate possible changes in size or shape, events that would require surgical intervention²¹.

The size of the defect size needs to be considered, because it represents a limitation, but not an impediment, when planning implant-supported rehabilitation and other interventions in the affected area. The recognition and definitive diagnosis of this condition are essential, particularly because of the increasing use of rehabilitation treatments with osseointegrated implants.

Multislice CT provides important information about the margins and content of suspicious areas. In the present case, this imaging technique demonstrated the high variability of tissues present in the defect and indicated their anatomic relationships. Although surgery can reduce bone thickness in the middle and inferior portion of the mandible, it is unnecessary in most cases of Stafne bone defects. However, periodic radiographic follow-up is recommended.

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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.
- Çağlayan F, Tozoğlu U. Incidental findings in the maxillofacial region detected by cone beam CT. Diagn Interv Radiol. 2012;18:159-63.
- 3. Togan B, Gander T, Lanzer M, Martin R, Lübbers H. Incidence and frequency of nondental incidental findings on cone-beam computed tomography. J Craniomaxillofac Surg. 2016;44:1373-80.
- 4. 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.
- Minowa K, Inoue N, Sawamura T, Matsuda A, Totsuka Y, Nakamura M. Evaluation of static bone cavities with CT and MRI. Dentomaxillofac Radiol. 2003;32:2-7.
- 6. Campos PS, Panella J, Crusoé-Rebello IM, Azevedo RA, Pena N, Cunha T. Mandibular ramus-related Stafne's bone cavity. Dentomaxillofac Radiol. 2004;33:63-6.
- 7. Apruzzese D, Longoni S. Stafne cyst in an anterior location. J Oral Maxillofac Surg. 1999;57:333-8.
- Katz J, Chaushu G, Rotstein I. Stafne's bone cavity in the anterior mandible: a possible diagnostic challenge. J Endod. 2001;27:304-7.
- Ozaki H, Ishikawa S, Kitabatake K, Yusa K, Tachiaban H, Iino M. A Case of Simultaneous Unilateral Anterior and Posterior Stafne Bone Defects. Case Rep Dent. 2015;2015:983956.
- 10. Bayrak NB. A rare presentation of Stafne bone cyst. J Stomatol Oral Maxillofac. Surg. 2019. pii: S2468-7855(19)30050-3. [Epub ahead of print]
- 11. Shimizu M, Osa N, Okamura K, Yoshiura K. CT analysis of the Stafne's bone defects of the mandible. Dentomaxillofac Radiol. 2006;35:95-102.
- Taysi M, Ozden C, Cankaya B, Olgac V, Yıldırım S. Stafne bone defect in the anterior mandible. Dentomaxillofac Radiol. 2014;43:20140075.
- Turkoglu K, Orhan K. Stafne bone cavity in the anterior mandible. J Craniofac Surg. 2010;21:1769-75.
- 14. Sisman Y, Miloglu O, Sekerci AE, Yilmaz AB, Demirtas O, Tokmak TT. Radiographic evaluation on prevalence of Stafne bone defect: a study from two centres in Turkey. Dentomaxillofac Radiol. 2012;41:152-8.
- 15. Lee KH, Thiruchelvam JK, McDermott P. Unusual Presentation of Stafne Bone Cyst. J Maxillofac Oral Surg. 2015;14:841-4.

- 16. 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.
- 17. Flores Campos PS, Oliveira JA, Dantas JA, de Melo DP, Pena N, Santos LA, et al. Stafne's Defect with Buccal Cortical Expansion: A Case Report. Int J Dent. 2010;2010:515931.
- Segev Y, Puterman M, Bodner L. Stafne bone cavity–magnetic resonance imaging. Med Oral Patol Oral Cir Bucal. 2006;11:E345-7.
- Adisen M, Yılmaz S, Misirlioglu M, Atil F. Evaluation of volumetric measurements on CBCT images using Stafne bone cavities as an example. Med Oral Patol Oral Cir Bucal. 2015;20:e580-6.
- 20. Hisatomi M, Munhoz L, Asaumi J, Arita E. Stafne bone defects radiographic features in panoramic radiographs: Assessment of 91 cases. Med Oral Patol Oral Cir Bucal. 2019;24:e12-9.
- 21.Precht C, Stockmann P, Neukam FW, Schlegel KA. Enlargement of a Stafne cyst as an indication for surgical treatment - a case report. J Craniomaxillofac Surg. 2013;41:270-3.