

Stafne bone defect: a report of two cases and diagnostic considerations

Defeito ósseo de Stafne: relato de dois casos e considerações sobre o diagnóstico

Rodrigo Porpino MAFRA¹, Marcelo Gadelha VASCONCELOS², Rodrigo Gadelha VASCONCELOS², Emeline das Neves de Araújo LIMA³, Déborah Pitta Paraíso IGLESIAS⁴, Lélia Maria Guedes QUEIROZ⁵

1 Post-graduation Program in Oral Pathology – Department of Dentistry – Federal University of Rio Grande do Norte (UFRN) – Natal – RN – Brazil.

2 PhD in Oral Pathology – Department of Dentistry – State University of Paraíba (UEPB), Araruna – PB – Brazil.

3 Department of Dentistry – Federal University of Sergipe (UFS) – Lagarto – SE – Brazil.

4 Department of Pathology – Federal University of Pernambuco (UFPE) – Recife – PE – Brazil.

5 Post-graduation Program in Oral Pathology – Department of Dentistry – Federal University of Rio Grande do Norte (UFRN) – Natal – RN – Brazil.

ABSTRACT

Stafne bone defects are asymptomatic lingual bone depressions of the lower jaw, frequently caused by soft tissue inclusion. The common variant of this entity affects the third molar region, below mandibular canal, and is mostly diagnosed incidentally during routine radiographic examination. The uncommon anterior variant is relatively rare and located in the premolar region of the mandible. Sublingual salivary glands are thought to be responsible for the development of this variant. The aim of this report was to describe a case of Stafne bone defect in the anterior region of mandible and a case in posterior mandible, with emphasis on clinical and radiographic findings. Dental clinicians should be aware of this entity, aiming to avoid unnecessary biopsies. In most cases, clinical and radiographic follow-up is the recommended conduct.

KEYWORDS

Bone cysts; Case reports; Maxillofacial abnormalities.

RESUMO

Defeitos ósseos de Stafne são cavidades ósseas assintomáticas localizadas em mandíbula, frequentemente causadas pela inclusão de tecidos moles. A variante comum desta entidade acomete a região de terceiros molares, abaixo do canal mandibular, sendo geralmente diagnosticada de forma incidental durante exames radiográficos de rotina. A variante em região anterior é incomum e localiza-se nas proximidades dos pré-molares mandibulares. Acredita-se que as glândulas salivares sublinguais estejam implicadas no desenvolvimento desta variante. O objetivo deste relato foi descrever um caso de defeito ósseo de Stafne na região anterior de mandíbula e um caso em mandíbula posterior, com ênfase nos achados clínicos e radiográficos. Cirurgiões-dentistas deveriam ter conhecimento desta entidade para evitar biópsias desnecessárias. Na maioria dos casos, acompanhamento clínico-radiográfico constitui a conduta recomendada.

PALAVRAS-CHAVE

Anormalidades maxilofaciais; Cistos ósseos; Relatos de casos.

INTRODUCTION

Edward Stafne was the first to report the presence of “bone cavities” in the angle of the mandible, all of which were found near this location [1]. Stafne bone defects (SBDs) are non-progressive yet non-healing bone cavities situated frequently near the angle of the mandible, distal to third molar and caudal to the inferior alveolar nerve [2]. Rare locations include the anterior mandible and the ramus. In most instances, salivary gland tissue can be found in these defects, which are of unclear pathogenesis [2,3].

This entity presents as asymptomatic lingual bone depressions of the lower jaw that are frequently caused by soft tissue inclusion. The anterior variant of a SBD is relatively uncommon and usually located in the premolar region of the mandible. Sublingual salivary glands are thought to be responsible for this variant. However, other structures such as lymphoid, connective, muscular or vascular tissues might be associated with the anterior variant of SBD [4].

The prevalence of SBDs is approximately 0.10% to 0.48% on image examinations [3,5,6]. The diagnosis has usually relied on imaging studies, and panoramic radiographies are used for regular examination [5]. This entity presents a radiolucent ovoid shadow with sclerotic borders under mandibular canal, usually located at the angle of the mandible, but may also be found at the anterior region or other areas of the mandible [3,4]. Definitive diagnosis relies on biopsy which is not always necessary. Computed tomography, magnetic resonance imaging and sialography of the submandibular gland may provide sufficient information to provide a diagnosis [7] and the treatment is essentially clinical follow-up [8].

The aim of this article is to report two different cases of SBD, emphasizing clinical and radiographic aspects relevant for the diagnosis and monitoring of this pathology.

CASE REPORTS

Case report 1

A 32-year-old white man attended an Oral Diagnosis Service, complaining of a lesion in the anterior portion of the left mandible, from canine to first premolar, with two years of evolution. The patient’s medical history was non-contributory and no palpable cervical lymph nodes were present. Intraoral examination revealed the presence of a clinically palpable cavity or depression, rounded with irregular surface, color similar to oral mucosa, firm and asymptomatic, measuring 01 cm in greatest diameter (Figure 1). All the teeth in that region responded within normal limits to pulp vitality tests.

Panoramic and periapical radiographies confirmed the presence of a well-defined radiolucent lesion with sclerotic margins in the anterior mandible, associated with the sublingual gland, mimicking a periapical lesion (Figure 2). Thereby, there was no need for biopsy due to clinical and radiographic features of the SBD. A two-year follow-up ensued. No dimensional changes were observed on the panoramic radiograph. Therefore, it was confirmed the diagnosis of SBD.

Case report 2

A 73-year-old white man attended an Oral Diagnosis Service for routine radiographic examination. The patient’s medical history was non-contributory. Extraoral examination revealed no remarkable features or palpable cervical lymph nodes. Intraoral inspection also showed no noteworthy findings, and all teeth in the region responded normally to the pulp vitality tests.

Radiographic examination confirmed the presence of a unilocular, well-circumscribed radiolucent lesion with sclerotic borders in the posterior mandible, between molars and mandibular angle, resembling a residual cyst (Figure 3). However, in view of the radiological

characteristics consistent with SBD, we opted for the follow-up of the patient. After 18 months, panoramic radiograph revealed no alterations in the ovoid radiolucency. Thus, it was established the clinical diagnosis of SBD.

DISCUSSION

Bone defects in the mandible have been described using different terms since Stafne, in 1942, reported on 35 radiolucent lesions in the mandibular angle [1]. Among the terms used to describe this entity, we can exemplify: lingual mandibular bone cavity, concavity, defect, or depression; Stafne defect, cavity or cyst; static or latent or idiopathic defect, cavity, or cyst; mandibular salivary gland inclusion; aberrant or ectopic salivary gland [9].

The pathogenesis of SBD is not clearly elucidated. There has been some debate as to

whether this entity is congenital or developmental in nature [5]. Normal submandibular salivary gland tissue is the most common histologic



Figure 1 - Lingual view of anterior mandible showing a concavity.



Figure 2 - a) Periapical radiograph; b) panoramic radiograph, showing the oval anterior mandibular bone defect.



Figure 3 - a) Panoramic radiograph; b) lateral radiograph of face, showing a mandibular unilocular radiolucency (arrows) located above inferior dental canal.

finding [3], suggesting its involvement in the origin of this defect. In a few cases, muscle, blood vessels, fat, fibrous connective tissue, or lymphoid tissue have also been reported [2,6,10].

The clinical findings of the current case 2 are in accordance with previous case series that have described epidemiological and clinical aspects of SBDs. The typical location of Stafne defects is between the mandibular canal and the angle of the mandible [4]. The anterior lingual variant, 7 times less frequent than the posterior, is usually located between the incisor and premolar areas, above the insertion of the mylohyoid muscle. Variants with lingual or buccal involvement of the ascending ramus of the mandible are very unusual findings [8].

The anterior variant of a SBD is considered to be rare. In contradistinction to posterior lesions, anterior lingual mandibular SBDs may be difficult to diagnose. They are found between or below the roots, superimposed over the roots [6], as in our case 1, or at sites of previous extraction.

According to Philipsen et al. [8], SBDs are much more frequent in males, with a male to female ratio of 6:1. Patients are usually diagnosed between 50 and 70 years of age [2]. The characteristics of our patients are similar to those described in previous reports [2,3,5,9].

Most of the SBDs were discovered during routine oral radiographic examination because they were usually asymptomatic and nonprogressive. The lingual defect rarely can be clinically palpated [5]. In our case 1, we related an unusual presentation in the anterior location, detected during routine oral examination. The radiographic findings, observed in the case 2 mentioned above, are typical on SBD. The defect appears as a circumscribed, unilocular osteolytic radiolucency under the mandibular canal, usually at the angle of the mandible, but can also occur at the anterior region or other areas of the mandible [3,4,6]. Sometimes, the

borders are sclerotic, and in other instances, they are not clearly defined [5].

Review of the literature reveals that radicular cyst and residual cyst, as in our case 1 and 2 respectively, as well as non-inflammatory odontogenic cysts, were the most frequent hypotheses considered in the differential diagnosis [10]. In a minority of cases, the differential diagnosis may include simple bone cyst, focal osteoporotic bone marrow defect, giant cell lesion, ameloblastoma and keratocystic odontogenic tumor [2,4,9]. Regarding the cases reported in this article, the findings in mandibular bone were diagnosed as SBDs mainly because of the location and radiographic appearance, but also due to clinical features such as absence of symptoms.

No treatment is necessary for SBDs in either posterior or anterior variants. Surgical exploration and biopsy should be performed only to rule out other pathological entities in atypical cases when the diagnosis is uncertain, or exceptionally when an additional pathology is suspected to develop in the entrapped salivary tissue [6]. Therefore, radiographic follow-up is recommended [4]. Miloğlu et al. [9] suggest that non-invasive radiographic examinations should be performed for diagnosis and monitoring of these patients, aiming to avoid unnecessary surgical approaches.

We emphasize that clinicians should be aware of clinical and radiographic aspects of SBDs and include it in the differential diagnosis of entities with similar features. The findings of SBDs are not of pathologic significance; they are generally regarded as innocuous and asymptomatic. Therefore, it is usually agreed that surgical treatment is not indicated, but clinical and radiographic follow-up should be conducted.

REFERENCES

1. Stafne EC. Bone cavities situated near the angle of the mandible. *J Am Dent Assoc.* 1942;29(17):1969-72.
2. Schneider T, Filo K, Locher MC, et al. Stafne bone cavities: systematic algorithm for diagnosis derived from retrospective data

- over a 5-year period. *Br J Oral Maxillofac Surg.* 2014 Apr;52(4):369-74. doi: 10.1016/j.bjoms.2014.01.017.
3. Dereci O, Duran S. Intraorally exposed anterior Stafne bone defect: a case report. *Oral Surg Oral Med Oral Pathol Oral Radiol.* 2012 May;113(5):e1-3. doi: 10.1016/j.tripleo.2011.07.029.
 4. Herranz-Aparicio J, Figueiredo R, Gay-Escoda C. Stafne's bone cavity: An unusual case with involvement of the buccal and lingual mandibular plates. *J Clin Exp Dent.* 2014 Feb;6(1):e96-9. doi: 10.4317/jced.51229.
 5. Queiroz LM, Rocha RS, de Medeiros KB, da Silveira EJ, Lins RD. Anterior bilateral presentation of Stafne defect: an unusual case report. *J Oral Maxillofac Surg.* 2004 May;62(5):613-5.
 6. Ozaki H, Ishikawa S, Kitabatake K, Yusa K, Tachibana H, Iino M. A Case of Simultaneous Unilateral Anterior and Posterior Stafne Bone Defects. *Case Rep Dent.* 2015. doi: 10.1155/2015/983956.
 7. Shimizu M, Osa N, Okamura K, Yoshiura K. CT analysis of the Stafne's bone defects of the mandible. *Dentomaxillofac Radiol.* 2006 Mar;35(2):95-102. doi: 10.1259/dmfr/71115878.
 8. Philipsen HP, Takata T, Reichart P, Sato S, Swei 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 Sep;31(5):281-90. doi: 10.1038/sj.dmfr.4600718.
 9. Milošlu Ö, Sekerci AE, Yasa Y, Dagistan S. Unilateral bone cavities situated near the angle of the mandibula. *J Craniofac Surg.* 2015 Jan;26(1):e27-8. doi: 10.1097/SCS.0000000000001262.
 10. Katz J, Chaushu G, Rotstein I. Stafne's bone cavity in the anterior mandible: a possible diagnostic challenge. *J Endod.* 2001 Apr;27(4):304-7. doi: 10.1097/00004770-200104000-00020.

Rodrigo Porpino Mafra
(Corresponding address)

Av. Senador Salgado Filho, 1787, CEP: 59056-000, Lagoa Nova,
Natal, RN, Brazil.
Phone/Fax: 55-84-3215-4108 or 55-84-3215-4138
E-mail: rodrigo_p.m@hotmail.com

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