

NATHÁLIA RODRIGUES GOMES

**DEFEITO ÓSSEO DE STAFNE EM REGIÃO ANTERIOR ASSOCIADO
A REABSORÇÃO RADICULAR EXTERNA: *RELATO DE UM CASO*
*RARO***

Faculdade de Odontologia
Universidade Federal de Minas Gerais
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Monografia apresentada ao Curso de Especialização Radiologia Odontológica e Imaginologia da Faculdade de Odontologia da Universidade Federal de Minas Gerais, como requisito parcial à obtenção do título de Especialista em Radiologia Odontológica e Imaginologia.

Orientadora: Profa. Dra. Cláudia Brasileiro Borges

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Ata da Comissão Examinadora para julgamento de Monografia da aluna **NATHÁLIA RODRIGUES GOMES**, do Curso de Especialização em Radiologia Odontológica e Imaginologia, realizado no período de 02/03/2020 a 07/11/2020. Aos 27 dias do mês de outubro de 2020, às 16:00 horas, por meio da plataforma virtual Microsoft Teams®, reuniu-se a Comissão Examinadora, composta pelos professores Cláudia Borges Brasileiro (orientadora), Luciana Cardoso Fonseca Terzis e Maurício Augusto Aquino de Castro. Em sessão pública foram iniciados os trabalhos relativos à Apresentação da Monografia intitulada “**Defeito Ósseo de Stafne em região anterior associado à reabsorção radicular externa: relato de um caso raro**”. Terminadas as arguições, passou-se à apuração final. A nota obtida pela aluna foi 100,0 (cem) pontos, e a Comissão Examinadora decidiu pela sua **APROVAÇÃO**. Para constar, eu, Cláudia Borges Brasileiro, Presidente da Comissão, lavrei a presente ata que assino, juntamente com os outros membros da Comissão Examinadora. Belo Horizonte, 27 de outubro de 2020.

Prof. Cláudia Borges Brasileiro
Orientadora

Prof. Luciana Cardoso Fonseca Terzis

Prof. Maurício Augusto Aquino de Castro

RESUMO

O defeito ósseo de Stafne (DOF) é incomum e normalmente se apresenta como uma depressão óssea lingual assintomática que acomete a região de ângulo da mandíbula, abaixo do canal mandibular. Devido sua rara localização anterior este relato descreve o caso de uma paciente mulher de 37 anos com imagens radiográficas que revelam uma imagem unilocular radiolúcida e bem definida localizada abaixo dos ápices dos incisivos e canino inferior esquerdo e adjacente a raiz do primeiro pré-molar, com aspecto sugestivo de reabsorção da raiz do canino. O exame intraoral mostrou dentes com ausência de lesões cáries e positivos ao teste de vitalidade pulpar. Tomografia computadorizada de feixe cônico revelou invaginação da cortical lingual, lesão bem corticalizada e que se estende do primeiro pré-molar ao incisivo central inferiores do lado esquerdo. O ápice do incisivo lateral apresenta discreta reabsorção radicular e o canino apresenta severa reabsorção radicular externa. Uma biópsia incisional foi realizada e o exame microscópico demonstrou a presença de tecido glandular salivar e o diagnóstico foi de defeito ósseo de Stafne em região anterior associado a reabsorção radicular externa e este relato discute as hipóteses para patogênese desse achado único.

Palavras-chave: Defeito ósseo de Stafne. Reabsorção radicular. Radiografia panorâmica. Tomografia computadorizada de feixe cônico.

ABSTRACT

Anterior Stafne bone defect associated with external root resorption: an unusual presentation

Stafne bone defect (SBD) is an uncommon lingual bone depression usually asymptomatic that often occur near the angle of the mandible below the mandibular canal. Due to the rare anterior localization this article describes a 37-years-old female with radiographic images that revealed well-defined unilocular radiolucent lesion below the apices of the lower left incisors, canine and adjacent to first premolar, with root resorption of the canine. Intraoral examination showed non-carious teeth and positive vitality tests. Cone beam computed tomography revealed an invagination of the lingual cortex, well corticated, which reaches the buccal cortex, extending from first premolar to central incisor tooth in the inferior left side. Lateral incisor presented mild apical resorption while canine tooth presented severe root resorption. Incisional biopsy was performed and microscopic examination showed the presence of salivary gland tissue. The diagnosis was anterior SBD associated with external apical root resorption and the article discuss the hypothesis for the pathogenesis of this unique finding.

Keywords: Stafne bone defect. Root resorption. Panoramic radiography. Cone beam computed tomography.

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1 CONSIDERAÇÕES INICIAIS

O defeito ósseo de Stafne (DOS) foi primeiramente descrito por Edward Stafne, em 1942, como uma imagem radiolúcida bem definida, redonda ou ovoide na região posterior da mandíbula, próximo ao ângulo da mandíbula, encontrados como um achado radiográfico e que normalmente não requer cirurgia (STAFNE, 1942). Outros termos como cavidade óssea estática, cisto ósseo estático ou latente, cavidade óssea lingual mandibular, defeito da mandíbula também se referem a essa entidade.

Foi demonstrada uma prevalência de 0,009 a 0,48% do DOS, com homens representando a maioria dos casos com uma taxa de 3:1 (PHILIPSEN, TAKATA, *et al.*, 2002, TURKOGLU, SURGERY, *et al.*, 2010). A maioria dos casos se apresenta unilateralmente e na região posterior da mandíbula, especialmente entre o primeiro molar e o ângulo da mandíbula (TURKOGLU, SURGERY, *et al.*, 2010) e o primeiro relato de um DOS em região anterior foi relatado em 1957 (ALTWAIM, AL-SADHAN, 2019, BORNSTEIN, WIEST, *et al.*, 2009)

DOS é geralmente encontrado como um achado radiográfico. Uma radiografia panorâmica é adequada para o diagnóstico de defeitos localizados na região posterior mandibular devido sua aparência patognomônica. Entretanto, a avaliação de defeitos ósseos anteriores pode ficar comprometida por esse tipo de exame (DERECI, DURAN, 2012). Técnicas mais avançadas de imagem, como a tomografia computadorizada (TC) e a ressonância magnética (RM), podem ser benéficas no diagnóstico de defeitos anteriores e para se saber a extensão de defeitos posteriores (GRIFFA, ZAVATTERO, *et al.*, 2014, HE, WANG, *et al.*, 2019, KATZ, 2001). Testes adicionais, como o de sensibilidade pulpar, podem ser necessários em situações especiais, para se evitar tratamento endodôntico desnecessário (DE COURTEN, KÜFFER, *et al.*, 2002, KATZ, 2001, TURKOGLU, ORHAN, 2010).

Portanto, esse relato se trata de um caso raro de DOS em região anterior, associado a reabsorção radicular externa em uma mulher de 37 anos e discute a literatura em relação a esta entidade.

2 OBJETIVOS

2.1 Objetivo geral

Apresentar um caso raro de defeito ósseo de Stafne em região anterior de mandíbula associado a reabsorção radicular.

2.2 Objetivos específicos

Revisão de literatura sobre casos de defeito ósseo de Stafne em região anterior de mandíbula.

Discutir a patogênese da reabsorção radicular associada ao defeito ósseo de Stafne.

3 RESULTADOS

Os resultados foram escritos em forma de artigo na língua inglesa e será submetido ao periódico internacional Dentomaxillofacial Radiology.

3.1 Artigo

TITLE: Anterior Stafne bone defect associated with external root resorption: an unusual presentation

KEYWORDS: Stafne bone defect, tooth resorption, panoramic radiography, cone beam computed tomography

INTRODUCTION

Stafne bone defect (SBD) was first described by Edward Stafne, in 1942, as a well-circumscribed, round or ovoid radiolucency in the posterior region of the mandible, near the angle, seen on radiographs that commonly do not require surgery. (1) Other terms like static bone cavity, static or latent bone cyst, lingual mandibular bone cavity, defect of the mandible, developmental lingual mandibular salivary gland depression are also referred to this entity.

A prevalence rate of SBD between 0.009 and 0.48% have been reported, with men representing most cases with a 3:1 ratio. (2, 3). The majority of cases of SBD is presented unilaterally and is located in the posterior region of mandible, especially between the first molar and the angle of the mandible (3) and the first report of an anterior SBD was made in 1957. (4,5)

SBD is seen as a radiographic finding for other purposes. A single panoramic radiograph is adequate for the diagnosis of posterior Stafne defects, because its pathognomonic appearance. However, evaluation of anterior defects may be compromised by this type of radiograph. (6) Improved imaging techniques, such as computerized tomography (CT) and magnetic resonance imaging (MRI), may be beneficial in the diagnosis of anterior defects and the extension of posterior defects. (7-9) Additional tests such as sensibility pulpal tests could be necessary in special situations, in order to avoid unnecessary endodontic therapy. (3, 8, 10)

Therefore, this paper aims to report an unusual case of anterior SBD, associated with external root resorption in a 37-year-old female and further discuss the literature concerning this entity.

CASE REPORT

A 37-year-old female patient presented to the Clinic of Pathology, Stomatology, and Radiology of the School of Dentistry, Universidade Federal de Minas Gerais (Belo Horizonte, Minas Gerais state, Brazil) referred by another professional for the evaluation of an unusual image observed in a routine radiography exam. During anamnesis, the patient reported to present arterial hypertension, which is usually controlled, and gastroesophageal reflux.

Clinical evaluation showed no signs of swelling, redness or inflammation in the soft tissue. Teeth #32, #33, #34 and #35 responses to both cold (Endo Ice, Maquira, Maringá, Brazil) and electricpult testing (Pulp Tester Digital VCR-200, Odous De Deus, Belo Horizonte, Brazil) were positive and similar to contra-lateral homologous.

The radiographic periapical and panoramic exam, brought by the patient, revealed a great unilocular, well-defined radiolucent area with lobulated aspect, partially corticalized, between lower left canine and first premolar, and an anomalous external root resorption of left lower canine (FIGURE 1).



FIGURE 1: A) Panoramic radiograph showing an unilocular radiolucent image between the roots of left lower incisors, canine and adjacent to first pre-molar root. B and C) Periapical images evidences the root resorption of left lower canine.

A cone beam computed tomography (CBCT) was requested in order to clarify the dimensions and limits of the lesion (FIGURE 2). The CBCT images revealed an osseous depression in the lingual region, extending to the buccal cortex, without breaking it, well limited, and promoting root resorption of the lingual medium and apical thirds of the left lower canine root and a mild apical resorption of the lateral incisor, extending from first premolar to central incisor tooth.

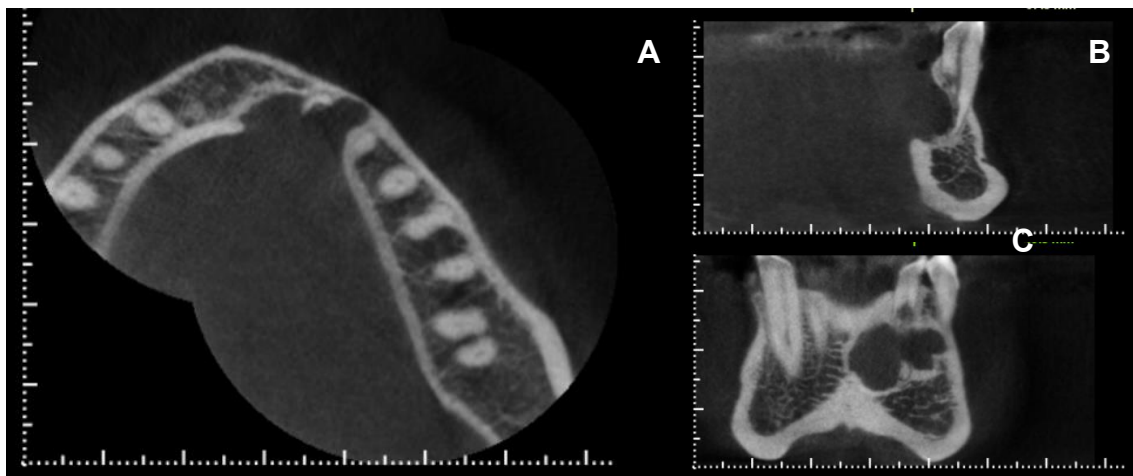


FIGURE 2: A) axial, B) sagittal and C) coronal slices of the CBCT exam show the lingual depression and left lower canine root resorption.

Considering the uncertainty about the diagnosis it was proposed to the patient an incisional biopsy aiming histological information for a definitive diagnosis. The removed fragment was sent to the pathology laboratory for further investigation. The patient was re-evaluated seven days after the surgical intervention and her recovery was uneventful.

The optical microscopic examination revealed the presence of several seromucous salivary gland lobes. In addition, secretory ducts, connective tissue and chronic inflammatory cells completed the histological findings. Thus, a definitive diagnosis of Stafne bone defect could be confirmed (FIGURE 4).

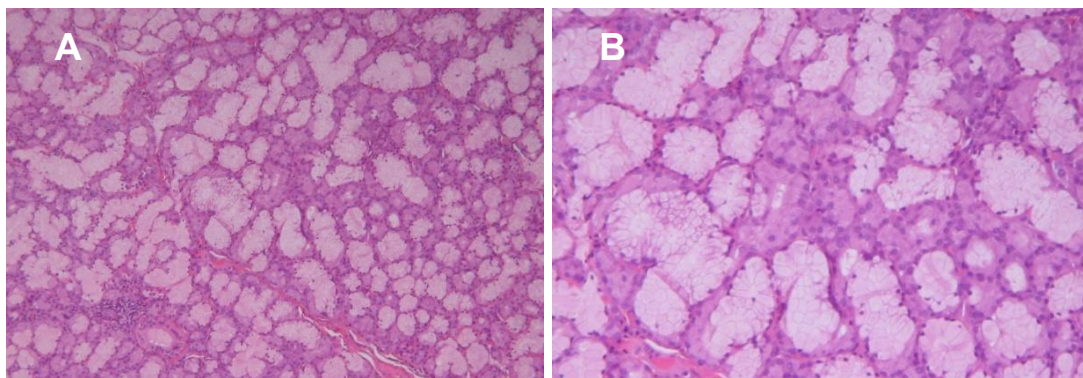


FIGURE 3: Histopathological examination of the fragment collected in the biopsy (Hematoxylin and eosin staining) A) original magnification X25 and B) original magnification X40 reveals salivary gland fragments with usual characteristics.

The patient is maintained in follow up, now of 24 months after the diagnosis achievement, without complaints and with no visible alterations.

DISCUSSION

SBD is a relatively rare, but well described, defect of the jaw bone, usually asymptomatic and nonprogressive, most often located in the posterior region of mandible of male adult patients. (1, 3, 10) The present article described a conservative approach for an unusual case of SBD, diagnosed in the anterior mandible of a female, with tooth resorption and tissue entrapment.

Pathogenesis of SBD is still not clear and there are some hypotheses. (6, 11) Some consider that during the mandibular bone developmental some part of the salivary gland tissue could stay entrapped inside the bone. (1) Others believe that this is a congenital cavity or condition of the development. (10) There is also the theory about the surrounding tissues making pressure over the salivary gland tissue. (2, 10) The present case lead the developmental theory as most suitable, because

histopathological findings revealed salivary gland tissue originating from the sublingual gland.

Differential diagnosis of anterior cases of SBD are difficult due to the radiographic aspect of a well-defined unilocular radiolucency superimposed to teeth roots, mimicking other unilocular lesions as odontogenic cysts, salivary gland tumors, fibro-osseous lesions and benign neurogenic tumors. (3, 11, 12) In the present case, differential diagnosis with apical periodontitis of endodontic origin was more challenging due to the presence of external root resorption in lower left canine. Nonetheless, clinical evaluation and the positive response to pulpal sensibility tests ruled out the possibility of periapical periodontitis of endodontic infection origin. (3, 8, 10) The external root resorption observed in the lower left canine is an intriguing finding and, as far as we are concerned, this is the first report of such a case in dental literature. Except for the resorption of primary shedding teeth, dental hard tissues are not normally resorbed and external root resorption (EER) invariably results from injury to periodontal ligament and root surface (13).

Therefore, ERR is a frequent sequel of trauma, inflammatory response to chronic endodontic infections, excessive pressure during orthodontic tooth movement, ectopic tooth eruption and the presence of granulomas, cysts and tumors. (14) The mechanisms behind the root resorption observed in the present case are essentially unknown since there is no paragon in dental literature. However, the histopathological findings revealed the presence of several seromucous salivary gland lobes, that are known to secrete not only digestive enzymes but also various cell growth factors, among them epithelial growth factor (EGF). EGF has been implicated in osteoclastogenesis and disease progression in giant cell tumor of bone. (15) In addition, the presence of the pericoronal follicle, rich in EGF, is responsible for the resorption of the surrounding bone tissue during permanent tooth eruption and accelerates primary root resorption. Finally, transient external root resorption in maxillary lateral incisor is the most common sequel of impaction and eruption, ectopic or not, of permanent canines or due to the pressure of eruption. (16) Therefore, it can be speculated that the same factors (pressure and production of EGF) might have participated in the etiopathogeny of the present external resorption associated with SBD. It is noteworthy the endodontic conservative approach adopted, since the present case of ERR was not originated or supported by infection inside the root canal. (14)

Most of the reported cases of anterior SBD cited were located between first molars and canines, thus just a few lesions were at the incisors region. (2, 8, 10) In the reported case, the defect was found between the apices of the mandibular left lateral incisor and left canine and its radiological presentation leads to uncertainties, even though SBD was considered as a diagnostic hypothesis.

When located at the incisors area the diagnosis can be more challenging. (3, 6) Usually radiographic evaluation is enough to achieve a diagnosis of SBD, but other exams as CT and MRI could be necessary for additional information. (8, 12) Dereci *et al.* presented a unique case of a SBD above mylohyoid muscle and Philipsen *et al.* related that the SBDs are commonly below the mylohyoid muscle. (2, 6)

Once the clinical and complementary examination are complete and the diagnosis can still not be elucidated, a surgical exploration with removal of fragment for histological analysis can be necessary. (6, 12) The histological exam result in the present study was fundamental to close the diagnosis and for the explaining and hypothesis of the tooth resorption.

The most appropriate treatment is the follow-up (7) as the surgical interventions are reserved for diagnostic purposes. (4, 7, 10) In the presented case, the indication of the incisional biopsy aimed to ensure the diagnosis, due to its uncommon clinical and radiological presentations, mainly regarding the entrapment of the muscular tissue into the mandibular body and the external root resorption of the adjacent canine albeit vital. However, periodic clinical examination and plain radiograph are considered enough to monitor the vast majority of the lesions. (3) From now, the follow-up of the reported patient can be done through clinical evaluation and evaluation of plan radiographs, in order to evaluate the process of the canine radicular resorption.

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4 CONSIDERAÇÕES FINAIS

O presente estudo descreve um caso raro de defeito ósseo de Stafne em região anterior de mandíbula promovendo reabsorção radicular. Apesar da patogênese desconhecida, o caso apresentado nos leva a considerar a hipótese que durante o desenvolvimento mandibular uma parte do tecido glandular salivar pode ter ficado atrelado ao interior do osso.

A reabsorção radicular encontrada no canino inferior do lado direito é o aspecto mais intrigante do caso apresentado, já que não há descrito na literatura outros casos de DOS com esse tipo de achado. Foi especulado que fatores como pressão do tecido glandular e produção de EGF (fator de crescimento epitelial) possam ter participado na etiopatogenia da reabsorção radicular externa associada ao DOS.

O relato do caso ressalta a importância da avaliação clínica e radiográfica na elaboração de diagnósticos diferenciais de casos atípicos, ressaltando-se também a importância do exame histopatológico para conclusão do referido caso.

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