CASE REPORT

Anterior lingual mandibular bone cavity: a case report

Cavidade óssea mandibular lingual anterior: um relato de caso

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ABSTRACT

Stafne’s bone cavity (SBC) is an asymptomatic lingual bone cavity situated near the angle of the mandible. The anterior variant of SBC, which shows a radiolucent unilateral ovoid lingual bone concavity in the canine-premolar mandibular region, is uncommon. A 73-year-old man was referred for assessment of loss of mandibular bone. Panoramic radiographs and computerized tomography scans showed a well-defined lingual bony defect in the anterior mandible. Analysis of imaginological documentation, made 14 years ago, revealed a progressive increase in mesiodistal diameter and intraosseous bony defect. The soft tissue obtained within the bony defect, microscopically revealed fibrous stroma containing blood vessels of varied caliber. The current anterior lingual mandibular bone defect case is probably caused by the salivary gland entrapped or pressure resorption, which can explain the SBC pathogenesis.

RESUMO

A cavidade óssea de Stafne (COS) é uma cavidade assintomática, localizada próximo ao ângulo da mandíbula, por lingual. A variante anterior do COS, a qual apresenta uma concavidade óssea lingual radiolúcida, ovoide e unilateral na região do canino-prémolar mandibular, é incomum. Um homem de 73 anos foi encaminhado para avaliação da perda óssea mandibular. A radiografia panorâmica e a tomografia computadorizada mostraram um defeito ósseo lingual bem definido na região anterior da mandíbula. A análise da documentação imaginológica, realizada há 14 anos, revelou um aumento progressivo do diâmetro mesiodistal e defeito ósseo intraósseo. A biópsia do tecido mole obtido do defeito ósseo revelou microscópicamente estroma fibroso contendo vasos sanguíneos de calibre variado. O presente caso de defeito ósseo mandibular na região lingual anterior é provavelmente causado por glândula salivar aprisionada ou reabsorção por pressão, o que pode explicar a patogênese da COS.

KEYWORDS

Bone defect; Mandible; Cone beam computed tomography; Diagnosis; Case report.

PALAVRAS-CHAVE

Defeito ósseo; Madibula, tomografia de feixe cônico; Diagnostico; Relato de caso.
INTRODUCTION

Stafne’s bone cavity (SBC) is an uncommon anomaly (prevalence of 0.08% [1] - 0.7% [2]) usually found in patients in the 5th or 6th decade of life and predominantly in males [1-4]. SBC has been described as an asymptomatic, well-defined radiolucent lingual bone cavity, mostly located in the posterior region of the mandible, below the inferior alveolar canal. The anterior variant of SBC, is uncommon [5], and often shows a radiolucent unilateral ovoid lingual bone concavity in the canine-premolar mandibular region, but bilateral variant has also been reported [6-8]. To the best of our knowledge, about 62 cases of SBC in the anterior mandible have been reported in the English-language literature (Table I). Moreover, unusual SBCs have been reported, including those cases with involvement of both lingual and buccal mandibular plates producing a tunnel-like lesion [9], location in the ramus of the mandible [10,11], simultaneous unilateral anterior and posterior bony defect [12], atypical trilobate aspect [13] or lesions completely occupied by fatty tissue [14].

Etiopathogenesis of the SBC is not fully understood. Although not all compatible, embryological origin and vascular alterations have been proposed as a possible cause of SBC. Currently, bone resorption in response to pressure of salivary gland tissue is the most widely accepted hypothesis [1,3,4,15], evidencing a peripheral origin [4]. Nevertheless, due to these defects are more frequently diagnosed in adults than children, it is suggested that the development of SBC probably occurs after the ossification of the mandible [3]. Interestingly, Philipsen et al. (2002) [3] suggest that a progressive reduction in bone volume could make that these depressions are visible on conventional radiographs around 35-40 years of age.

The current case report describes an anterior lingual mandibular bone concavity (showing SBC features), with documentation over a period of 14 years. In addition, a list of anterior variant of SBC cases previously reported in the literature is presented.

CASE REPORT

A 73-year-old man was referred for assessing an asymptomatic anterior lingual mandibular bone defect. Medical history and extraoral examination were non-contributory. It was described a recent history of perforation of the lingual cortical bone, and whose interior was empty. He denied any history of trauma and bone surgical procedures in the lesional area. However, it is evident that tooth extractions were performed in areas close to the bony defect. Intraoral examination revealed a lingual mandibular bone cavity, which presented empty and covered with erythematous oral mucosa. The patient had no sensory or motor deficiency, and there was no pain or lymphadenopathy. Moreover, no periapical lesions of odontogenic origin, near bony defect were present. Repeated thermal pulp testing performed in non-endodontically treated teeth produced normal results. Cone beam computerized tomography (CBCT) showed on the right side of the mandible an ovoid-shaped hypodense lesion of approximately 1.2 cm (buccolingual width) x 3.8 cm (anteroposterior length), evidencing a mesiodistal diameter expansion, vestibular cortical thinning and lingual cortical bone destruction (Figure 1). Histopathological examination of the soft tissue obtained within the bony defect was performed, which revealed fibrous stroma containing blood vessels of varied caliber (Figure 2). The clinicopathological correlation favored a diagnosis of variant anterior of SBC. Interestingly, two panoramic radiographs, taken in 2002 and 2010, as well as CBCT taken in 2010, could be evaluated after review of patient’s record (Figure 3 and 4). Noteworthy, they showed a progressive unilocular radiolucent area located in the anterior region of the right mandible below the apices of the second premolar (tooth # 29) to the canine (tooth # 27). Additionally, root canal treated teeth (tooth # 29 and tooth # 28) were observed (Figure 3a) and subsequently the loss of tooth # 29. Notice at level of tooth # 28 a discreet widening of the apical periodontium without contact with the radiolucent area (Figure 3b). In fact, both panoramic radiographs (Figure 3), show that the radiolucent area was distant and unrelated to the tooth apices, and by comparative analysis, there was a progressive increase in mesiodistal diameter of the radiolucent area. The CBCT scans performed in 2010 showed a well-defined lingual bony defect in the anterior mandible with expansion of the lingual cortical bone (Figure 4).
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Figure 1 - Cone beam computerized tomography (CBCT) taken in 2015, showing an unilateral ovoid-shaped hypodense lesion with increased dimensions. Axial (a) and cross-sectional (b) views show the complete destruction of the lingual cortical bone.

Figure 2 - Microscopic examination of soft tissue lining obtained from the lingual bony defect, revealed numerous blood vessels of varying size, supported by loose connective tissue (a, x4; b, x40; H&E stain).
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Figure 3 - Panoramic radiography images revealing a unilateral bone defect in the anterior mandible. (a) Image taken in 2002, revealing a unilocular radiolucent lesion in the canine and premolar area of the right mandible. (b) Image taken in 2010, showing a progressive increase of the lesion in mesiodistal diameter.

Figure 4 - Cone beam computerized tomography (CBCT) taken in 2010, showing unilateral ovoid-shaped hypodense lesion expanding the lingual cortical bone, in cross-sectional (a) and axial (b) views.
DISCUSSION

Based on studies of contemporary and archaeological cases, Philipsen (2002) [3] has described four topographical variants of SBC: 1) located in lingual anterior mandibular body (incisor-canine-premolar area), above the mylohyoid muscle; 2) located posterior to the mandibular angle-first permanent molar area, below the mandibular canal; 3) located to the ascending, lingual mandibular ramus, posterior to the lingual foramen, below the neck of the condyle; and 4) located to the buccal aspects of the ascending mandibular ramus (extremely rare). In general, posterior variant of SBC occurs most frequently [2-4,15] showing an prevalence of approximately 0.081% while the prevalence in anterior variant is 0.003% [16].

The review (English language literature) shown in the Table 1, presents a clinical data based on 63 cases of lingual bone cavity in the anterior mandible (including the current case) between 1957 and 2019. According to the anterior variant of mostly affected male patients (76%) with age ranging from 18 [17] to 76 years [12] (mean age, 46 years). This variant is most frequently described as an oval radiolucent unilateral lesion. Radiographically, the anterior variant presents less frequently well-defined sclerotic borders than posterior variant [3]. This finding could also explain why the diagnosis of anterior variant of SBC not considered in the differential diagnosis.

Regarding the location of the anterior variant of SBC, both sides of the mandible with similar frequency are affected, and mainly the premolar – canine region. Less common, this variant can have bilateral presentation (07 cases in men and 04 cases in women), appearing as a radiolucent lesion in each side of the mandible [7,8,18,19] or as a single radiolucent lesion crossing the midline [20-22] that may extend in the whole body of the mandible [6, 17, 23]. Additionally, Ozaki et al. (2015) [12] have reported a SBC which presented simultaneously anterior and posterior unilateral location.

The pathogenesis of the SBC is still controversial. Originally, the embryologic theory [24] has argued that a mandibular ossification defect of Meckel's cartilage could be the cause of this entity. Likewise, an entrapment of salivary gland tissue during ossification of the mandible has been suggested [10]. However, these theories are being disregarded because SBC is observed mostly in patients aged over 40 years [4].

On the other hand, most authors have accepted the pressure resorption theory [1,3,4,15] and considered it as the main etiological factor for majority of cases [25]. According to this theory, due to a chronic inflammatory process, the salivary gland hypertrophy is capable of exerting enough pressure to cause bone resorption. Apparently, there would be a relationship between the bone cavity and its corresponding submandibular gland [3,25]. Thus, hypertrophic submandibular gland could be the cause of the posterior variant of SBC, whereas an aberrant anterior lobe of the sublingual gland could be the cause of the anterior variant of SBC [4]. The lymphocytic infiltration found in salivary gland tissue, as possible cause of hypertrophied salivary glands, has been suggested to corroborate the theory [8] reinforcing the peripheral origin [4]. However, in contrast to this theory, some SBC case reports have revealed the absence of salivary gland tissue within the bony defect [26], whereas other studies showed presence of adipose tissue and bony fragments inside the bone cavity [14,27,28]. Interestingly, only 2 cases of the anterior variant of SBC have been reported presenting each one of them, among others, connective tissue (case 4) or blood vessels (case 62) (Table 1). In the current case, fibrous stroma containing blood vessels of varied caliber was observed. It should be noted that the tissue obtained was part of the lining of an empty central cavity, highly consistent with a reactive nature, and different from a vascular malformation or neoplasm.
The most SBC cases have been described as an asymptomatic and non-progressive anomaly [5, 29, 30]. Therefore, non-invasive diagnosis techniques have been preconized, avoiding surgical interventions, which could be an unnecessary option in the management of SBC [31]. In the current case, additional histological examination was required for accurate diagnosis, due to their progressive growing pattern, observed during the patient’s follow-up.

The SBC usually deserves no treatment [14,32], but when performed, conservative treatment is recommended [6]. Management should be based on integrated clinical and imaginological approach [9,33] or even three-dimensional imaging to rule out other odontogenic and non-odontogenic lesions [34].

To the best of our knowledge, about 62 cases of SBC in the anterior mandible have been reported in the English-language literature (Table I).

Table I - “Stafne bone cavity cases in the anterior mandible”

<table>
<thead>
<tr>
<th>Case</th>
<th>Authors (Year)</th>
<th>Age/ Sex</th>
<th>Unilateral/ Bilateral</th>
<th>Site</th>
<th>Content</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Richard and Ziskind (1957) [35]</td>
<td>46/M</td>
<td>Unilateral</td>
<td>Left canine and left first premolar</td>
<td>SG</td>
</tr>
<tr>
<td>2</td>
<td>Araiche and Brode (1959) [36]</td>
<td>55/M</td>
<td>Unilateral</td>
<td>Right central incisor to right premolar (edentulous)</td>
<td>SG</td>
</tr>
<tr>
<td>3</td>
<td>Camilleri (1963) [20]</td>
<td>31F</td>
<td>Bilateral</td>
<td>Incisal region (edentulous)</td>
<td>SG</td>
</tr>
<tr>
<td>4</td>
<td>Bergenholtz and Persson (1963) [27]</td>
<td>50/M</td>
<td>Unilateral</td>
<td>Left canine and left first premolar</td>
<td>CT, BF</td>
</tr>
<tr>
<td>5</td>
<td>Friedman (1964) [21]</td>
<td>54/M</td>
<td>Bilateral</td>
<td>Right canine to left canine</td>
<td>SG</td>
</tr>
<tr>
<td>6</td>
<td>Palladino et al. (1965) [37]</td>
<td>40/M</td>
<td>Unilateral</td>
<td>Right canine and right first premolar</td>
<td>SG</td>
</tr>
<tr>
<td>7</td>
<td>Abramson (1969) [38]</td>
<td>26/F</td>
<td>Unilateral</td>
<td>Right canine (edentulous)</td>
<td>SG</td>
</tr>
<tr>
<td>8</td>
<td>Miller and Winnecker (1971) [22]</td>
<td>48/M</td>
<td>Bilateral</td>
<td>Between central incisors</td>
<td>SG</td>
</tr>
<tr>
<td>9</td>
<td>Malkin and Berg (1974) [39]</td>
<td>54/M</td>
<td>Bilateral</td>
<td>Right canine to left canine</td>
<td>SG</td>
</tr>
<tr>
<td>10</td>
<td>Forrest (1974) [40]</td>
<td>Lateral incisor and canine</td>
<td>SG</td>
<td>Right canine and right first premolar</td>
<td>SG</td>
</tr>
<tr>
<td>11</td>
<td>Stone and Pedersen (1977) [41]</td>
<td>Right canine</td>
<td>SG</td>
<td>Right canine (edentulous)</td>
<td>SG</td>
</tr>
<tr>
<td>12</td>
<td>Plezia (1977) [42]</td>
<td>31/M</td>
<td>Unilateral</td>
<td>Right premolars</td>
<td>SG</td>
</tr>
</tbody>
</table>
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**Case Authors (Year) Age/ Sex Unilateral/ Bilateral Site Content**

<table>
<thead>
<tr>
<th>Case</th>
<th>Authors (Year)</th>
<th>Age/ Sex</th>
<th>Unilateral/ Bilateral</th>
<th>Site</th>
<th>Content</th>
</tr>
</thead>
<tbody>
<tr>
<td>35</td>
<td>de Courten et al. (2002) [57]</td>
<td>42/M</td>
<td>Unilateral</td>
<td>Between left first premolar and left second molar (edentulous)</td>
<td>SG</td>
</tr>
<tr>
<td>36</td>
<td>Dorman and Pense (2002) [58]</td>
<td>45/F</td>
<td>Unilateral</td>
<td>Between right central incisor and right first premolar</td>
<td>SG</td>
</tr>
<tr>
<td>37</td>
<td>Phillips et al. (2003) [59]</td>
<td>NM</td>
<td>Bilateral</td>
<td>Incisors, canines, premolars</td>
<td>No biopsy</td>
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<tr>
<td>38</td>
<td>Phillips et al. (2003) [59]</td>
<td>NM</td>
<td>Bilateral</td>
<td>Incisors, canines, premolars</td>
<td>No biopsy</td>
</tr>
<tr>
<td>39</td>
<td>Phillips et al. (2003) [59]</td>
<td>NM</td>
<td>Bilateral</td>
<td>Incisors, canines, premolars</td>
<td>No biopsy</td>
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<tr>
<td>40</td>
<td>Phillips et al. (2003) [59]</td>
<td>NM</td>
<td>Bilateral</td>
<td>Incisors, canines, premolars</td>
<td>No biopsy</td>
</tr>
<tr>
<td>41</td>
<td>Phillips and Yates (2004) [60]</td>
<td>52/M</td>
<td>Unilateral</td>
<td>Right first premolar</td>
<td>No biopsy</td>
</tr>
<tr>
<td>42</td>
<td>Quezine et al. (2004) [61]</td>
<td>32/F</td>
<td>Bilateral</td>
<td>Right lateral incisor–canine and left lateral incisor–canine</td>
<td>SG</td>
</tr>
<tr>
<td>43</td>
<td>Belmonte-Caro et al. (2005) [62]</td>
<td>68/M</td>
<td>Unilateral</td>
<td>Left canine and left first premolar</td>
<td>SG</td>
</tr>
<tr>
<td>44</td>
<td>Solomon et al. (2006) [63]</td>
<td>55/F</td>
<td>Bilateral</td>
<td>Canine</td>
<td>No biopsy</td>
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<tr>
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<td>Herbozo Silva et al. (2006) [64]</td>
<td>33/M</td>
<td>Bilateral</td>
<td>From right first molar to left first molar</td>
<td>SG</td>
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<tr>
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<td>Naih et al. (2007) [65]</td>
<td>47/M</td>
<td>Unilateral</td>
<td>Left premolars</td>
<td>SG</td>
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<tr>
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<td>25/F</td>
<td>Bilateral</td>
<td>Canine and premolars</td>
<td>No biopsy</td>
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<td>Unilateral</td>
<td>Right canine to right second premolar</td>
<td>No biopsy</td>
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<td>Left lateral incisor and left canine</td>
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<td>52/M</td>
<td>Unilateral</td>
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<td>SG</td>
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<td>No biopsy</td>
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<td>62/F</td>
<td>Unilateral</td>
<td>Right canine and right premolar (edentulous)</td>
<td>No biopsy</td>
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<td>Kraft et al. (2010) [71]</td>
<td>46/M</td>
<td>Unilateral</td>
<td>Left lateral incisor to left first premolar</td>
<td>SG</td>
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<td>Voss et al. (2010) [72]</td>
<td>58/M</td>
<td>Unilateral</td>
<td>Between the right canine and the right first molar</td>
<td>SG</td>
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<tr>
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<td>Voss et al. (2010) [72]</td>
<td>50/F</td>
<td>Unilateral</td>
<td>Left canine</td>
<td>No biopsy</td>
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<tr>
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<td>Dereci and Duran (2012) [73]</td>
<td>46/M</td>
<td>Unilateral</td>
<td>Left canine and left first premolar</td>
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<td>Kim et al. (2014) [74]</td>
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<td>Canine and premolars in the right and left mandible.</td>
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<td>Taysi et al. (2014) [75]</td>
<td>56/M</td>
<td>Unilateral</td>
<td>Left canine and left premolars</td>
<td>SG</td>
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<tr>
<td>59</td>
<td>Griffa et al. (2014) [76]</td>
<td>71/M</td>
<td>Unilateral</td>
<td>Left lateral incisor to left first premolar</td>
<td>SG</td>
</tr>
</tbody>
</table>

### REFERENCES


### CONCLUSION

According to the clinical, imaginological and pathological findings, the current case suggests a Stafne’s bone cavity (SBC) of uncommon localization and progression.
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