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An Unilateral Basal Bone Defect of the Mandible Occupied by Fatty Tissue: Stafne's Cavity

REINHARD E. FRIEDRICH[1], JOZEF ZUSTIN[2], HANNA A. SCHEUER[3], ALEXANDRE T. ASSAF[1] and ALEXANDER GRÖBE[1]

[1]Oral and Maxillofacial Surgery, [2]Pathology, and [3]Orthodontics, Eppendorf University Hospital, University of Hamburg, Hamburg, Germany

Abstract. *The differential diagnosis of osseous jaw lesions includes numerous entities. A specific mandibular bone defect known as Stafne's cavity or cyst presents wirh a characteristic cyst-like lesion on radiographs of the jaw. Although the differential diagnosis from other jaw lesions is mandatory, this lesion does not usually require surgical therapy. Current theories concerning the pathogenesis of Stafne's cavity prefer bone depression as a consequence of constant pressure arising from an adjacent salivary gland. We present a case of a large Stafne's cavity eroding the basal mandibular cortex that was found accidentally on an orthopantogram taken for diagnosis of dental diseases. The patient noted a slight pain on pressure during physical investigation. The lesion was completely occupied by fatty tissue, as shown during surgical exploration and as revealed by histological investigation of the specimen. The theory of parenchymal pressure as the cause of mandibular bone depression appears to be inapplicable in the present case. The pathogenesis of Stafne's cavity is still obscure. Differential diagnosis of mandibular lesions is essential in adequate treatment planning.*

In 1942, Stafne described a distinct osseous cavity in close proximity to the mandibular angle of unknown origin (1). In the original report, in all cases but one, the lesion was a unilateral finding (1). The lesion appears to have distinct expansion, usually restricted to the lingual aspect of the bone, and always presents caudally of the mandibular canal (1-3). The defect is known by a plethora of provisional technical terms but is best known as Stafne's cavity or cyst (3, 4). Because the osseous defect is entirely devoid of cystic components, the term Stafne's 'cyst' can be considered a misnomer; however, the radiological appearance of the cavity on plain radiographs indicates the differential diagnosis from cystic mandibular lesions (3). Thus, Stafne's 'cavity' is regularly listed in textbooks of oral pathology, radiology and surgery in chapters dealing with developmental defects, cysts or tumours of the oral and maxillofacial region (3-7). In order to avoid the use of eponyms, an alternative term 'mandibular lingual bone depression' has been proposed (3), allowing for the inclusion of further lingual mandibular bone lesions into one pathogenetic concept (2). The lesion was regarded as a developmental defect (1, 4, 8). However, this classification was challenged by others, with particular reference as to whether the osseous lesions should be assessed as a defect or depression (9, 14), *i.e.*, whether the radiographic detection of the lesion becomes visible in a region of impaired bone growth or follows a certain loss of bone (10, 11). However, this strict difference in terminology has not always been reflected in the literature regarding Stafne's cavity (2).

Most radiological reports and anthropometric studies have shown that the bone loss occurs predominantly on the lingual surface of the mandible (2, 9-12). On plain radiographs, in particular on orthopantomograms, Stafne's cavity appears as a roundish to ovoid radiolucency, extending in its longest diameter almost parallel to the basal border of the bone (2, 13-16). Stafne's cavity is usually an incidental finding (4, 8, 14). Because the defect may considerably vary in size, the typical complete osseous boundary of the defect may be lost, resulting in a relatively large radiolucency that can occasionally interrupt the mandibular basal cortex (1, 5). This pattern of bone depression does not fit with the concept of a strictly lingual bone resorption (2). On the other hand, Stafne's cavity never corrodes the mandibular canal and thus appears to be a self-limiting osseous process (4).

Stafne's cavity is a radiological finding recorded in adults (2, 8). The most widely supported theory on pathogenesis of the defect refers to the assumed ability of major salivary

Correspondence to: Professor R.E. Friedrich, Oral and Maxillofacial Surgery, Eppendorf University Hospital, University of Hamburg, Martinistr. 52, 20246 Hamburg, Germany. Tel: +49 40741053259, e-mail: rfriedri@uke.de

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gland tissue to erode bone (17). Indeed, many surgical interventions have revealed salivary gland tissue in the cavity itself (2). Furthermore, radiographic studies have repeatedly shown the presence of parts of the major salivary gland within these lesions (18). Tissues other than salivary glands were also found following surgical exploration (16), including fat (19, 20) and empty spaces (21) but these reports were cast into doubt due to possible inadequate surgical exploration (2, 3). It must be mentioned that one report described the presence of pleomorphic adenoma at the site of a Stafne's cavity (22), thus, restricting a wait-and-see policy generally proposed by opinion leaders in the fields of oral pathology (2-4).

We present a case of an extensive Stafne's cavity filled with fat but devoid of salivary gland tissue, which raises the question whether the salivary gland tissue observed within this type of bone depression is actually a cause of this unique bony cavitation or is simply a bystander.

Case Report

An 80-year-old male patient was referred to our outpatient clinic for the treatment of an osteolytic lesion in the right mandibular angle that was depicted on an orthopantomogram taken for dental treatment planning (Figure 1A). On admission, the patient exhibited an intact oral mucosa and a partial dentition. The radiographic characteristics of the osteolysis of the mandible depicted on the radiograph were consistent with the tentative diagnosis of a bone defect as described by Stafne (1). The patient denied any history of trauma to the region of interest, or surgery, except for tooth extractions. Although the patient claimed to be sensitive to pressure in the basal portion of the mandible of the affected side, in particular in the palpable notch, this finding was not consistently repeatable during the physical investigation. The proprioceptive sensibility of the lips was symmetrically established and the lips responsed well to appropriate haptic stimuli. We decided to explore the region and define the soft tissue occupying the defect. Under general anaesthesia, the vestibular mucosa of the right side of the mandible was incised to the periosteum, then the periosteum was detached from the bone and the vestibular aspect of the bone was exposed from the angle to the mental foramen. Somewhat beyond the mandibular basal border, a cranially angulated defect was exposed that was completely filled with yellowish connective tissue, consistent with the typical macroscopic aspect of fat (Figure 1B). This tissue was excised after complete exploration of the cavity, including the lingual bony surface, and consecutive detachment of the connective tissue inside the defect from the lingual periosteum. Bleeding of the wound was immediately and successfully stopped, the oral wound was closed by primary intention, and the tissue was prepared for histological investigation. Subsequent healing was uneventful and the patient experienced no neurological deficit after surgery.

The histological investigation revealed fatty tissue with no evidence of salivary gland tissue. The biopsy of adjacent bone showed normal cortical formation and trabecular network with adhering muscle.

Discussion

This case report clearly shows the presence of fatty tissue in a mandibular bicortical defect affecting a large part of the bone caudal of the mandibular canal. The photo-documentation definitely shows the fatty tissue billowing from the cavity. No surgical procedure displaced salivary gland tissue (2, 3).

Localization, physical findings and radiological characteristics of the defect found here are in accordance with the definition of the lesion as originally described by Stafne (1). Extensive loss of the basal cortex was evident in the present case. This finding reinforced the indication for surgical intervention. Indeed, the typical finding in Stafne's cavity is bone depression of the lingual mandibular side that does not extend to the buccal cortex (2-5). On plain radiographs, this pattern of osseous defect results in a roundish radiolucent lesion, with clear circular borders on all sides. The developmental defect grows slowly, as seen in the marked radiopaque line that borders the defect in many cases, indicating the capability of the bone to delineate the lesion. However, it is also well-known that the extension of the defect may affect the basal cortex (17), resulting in a remarkable loss of basal cortical bone (5). The largest lesions were found in older patients (13).

It is unclear why this growth is directed caudally, also in the sagittal plane, and can even transverse the bone completely, but does not affect cranially-located structures, *i.e.* the mandibular canal (2-4). Indeed, this defining feature of this entity should alert the clinician not to address cystic lesions beyond the mandibular canal as Stafne's cavity (23). The proximity of the submandibular gland to the lingual aspect of the bone and the occasional evidence of salivary gland tissue inside the defect supported the concept of a pressure-induced bone depression. However, this concept does not explain how a healthy salivary gland can exert such localized pressure on bone so as to force bone remodelling. Interestingly, the schematic description of formal pathogenesis of Stafne's cavity in terms of radiological stages provided by Ariji *et al.* (18) and adopted by others (2, 3), shows no salivary gland tissue in the first three out of four stages of mandibular lingual bone depression. After development of a lingual bone defect, the cavity becomes occupied by salivary gland tissue *via* a non-declared mechanism (2, 3). It is not clear how these steps of the pathogenesis of the cavity are linked. Indeed, in the

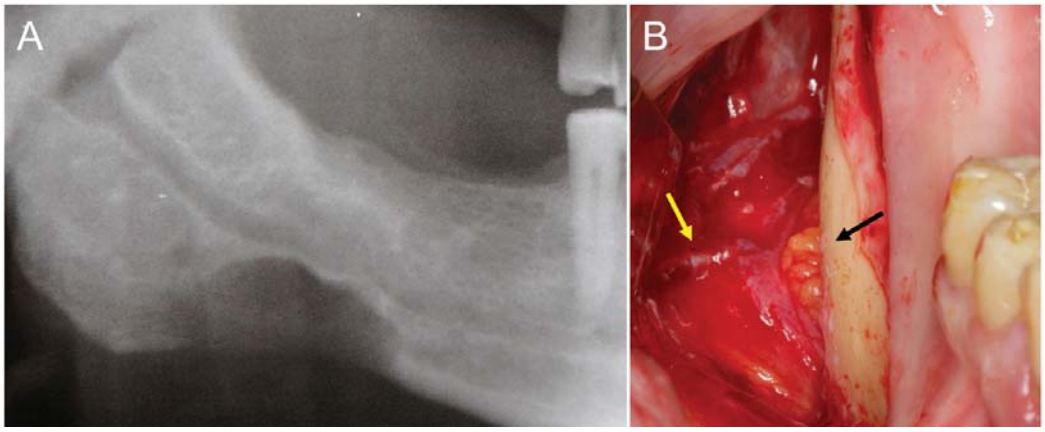

Figure 1. *A: Large ovally-shaped defect of the right mandibular base below the mandibular canal. B: This intraoperative image was taken after detachment of the buccal periosteum from the mandible and separation of the soft tissue occupying the mandibular defect. Yellowish fatty tissue can be seen to ooze out the cavity after incision of the periosteum (right arrow). Note the facial artery (left arrow) attached to the buccal soft tissue in close proximity to the defect (9).*

detailed description of computed-tomographic findings in Stafne's cavity, these authors described both parts of the salivary gland and exclusively fatty tissue in the cavities of the lingual aspect of the mandible (18). Therefore, it is currently not clear whether the salivary gland growth into the bone is simply a consequence of unclear bone resorption not related to the gland itself or indeed is caused by pressure of the gland. Our findings do not support the theory of glandular pressure on bone. Alternatively, a vascular pathogenesis was proposed to explain this entity (9). The twisted course of the facial artery around the basal border of the dorsal part of the mandibular corpus matched the extension of the defect in many cases (9). Alterations of the vessel, *e.g.* atherosclerosis or hypertension, may put pressure on the surrounding tissue resulting in bone resorption (9). Indeed, in the current case, the facial artery was running in close proximity to the defect (Figure 1B). However, the extension of the lesion was by far greater than the area covered by the vessel. Furthermore, this concept should be validated by more frequent bilateral findings, which is not the case.

Conclusion

Stafne's cavity is a well-known radiological finding of the mandible that usually deserves no treatment. However, extensive lesions may appear like tumourous osseous resorptions that should be adequately diagnosed. Thus, the surgical exploration cannot be avoided in every case that retrospectively may be diagnosed as a Stafne's cavity. The pathogenesis of the lesion is currently not known. The present report reveals the absence of salivary gland tissue from inside the lesion. Thus, the hypothesis of glands exerting pressure on bone followed by bone resorption is not supported in this case. The differential diagnosis of the entity is usually easily made on plain radiographs. In certain cases, in particular in extensive bicortical defects, surgical intervention is justified in order to exclude different pathologies that may require more advanced surgery.

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