



Stafne cyst in an 80 year old male: A case report

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ABSTRACT

The Stafne defect is thought to be a normal anatomical variant, as the depression is created by ectopic salivary gland tissue associated with the submandibular gland and does not represent a pathologic lesion as such. It is classed as a pseudo cyst, since there is no epithelial lining or fluid content. This defect is usually considered with other cysts of the jaws, since it can be mistaken for such on a radiograph. It is usually asymptomatic. Medical imaging such as traditional radiography or computed tomography is required to demonstrate the defect. Usually the defect is unilateral, but occasionally can be bilateral. We report a case of Stafne cyst in an 80 year old male.

Keywords: Stafne cyst, Idiopathic bone cavity, Static bone cyst

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INTRODUCTION

The Stafne bone defect was first described by Stafne in 1942. [1] Since then, numerous cases have been reported. The exact pathogenesis is still obscure. Stafne suggested that the cavity could result from a failure of normal bone deposition in the region formerly occupied by cartilage. Various explanations exist regarding the pathogenesis of Stafne cyst such as localized pressure atrophy of the lingual surface of mandible from the adjacent salivary gland, failure of normal bone deposition in the region previously occupied by Meckel's cartilage and development of mandible around entrapped submandibular salivary gland during the embryonic life, making the etiology of this condition not well defined. [2] Stafne bone defect has anterior and posterior variants. The posterior variant is the most known variant of the defect and is located between the mandibular angle and first mandibular molar tooth below the inferior dental canal. Variations such as lingual anterior variant, the medial ramus variant, bilateral presentation and bilocular defect, multilocular defect, or perforation of buccal cortical plate have been reported occasionally. Stafne bone defects were asymptomatic, with a predilection for men between 50 and 70 years. The diagnosis of this defect is incidental, since patients do not usually present clinical symptoms. In the orthopantomograph, the technique which usually first identifies this entity, a radiolucent image with a well-defined sclerotic border is generally observed. [3]

EPIDEMIOLOGY

The incidence is 0.1% - 6.06% in different reports. [4] The posterior lingual variant and the anterior lingual variant have an incidence of about 0.10-0.48 and 0.009%, respectively. [5] The age range is quite wide, but there is a peak incidence in the fifth-sixth decades with a male predominance. Stafne's defect is uncommon, and has been reported to develop anywhere between the ages of 11 and 30 years old, although the defect is developmental, it does not seem to be present from birth, implying that the lesion develops at a later age. Usually the defect is unilateral and most commonly occurs in men. [6]

CAUSES

The Stafne defect is thought to be caused by an ectopic portion of the submandibular salivary gland which causes the bone of the lingual cortical plate to remodel. Rarely, the defect can be completely surrounded by bone, and this has been theorized to be the result of entrapment of embryonic salivary gland tissue within the bone. Similar, but rarer, defects may be present in the anterior portion of the lingual surface of the mandible. These are not termed Stafne defects which specifically refers to

the posterior location. The anterior defects may be associated with the sublingual salivary gland. [7]

CASE REPORT

An 80 year old male patient reported to the Department of Oral & Medicine & Radiology, Institute of Dental Education and Advance Studies, Gwalior, Madhya Pradesh with a chief complaint of inability to open mouth since 2 months. Patient gave history of eventful extraction in lower left back jaw region 2 months back. Hence post extraction trismus gradually progresses. Intraoral examination revealed generalized attrition as well as recession.

Teeth present

15 14 13 12 11 21 22 23 24 25 26 27 28
48 47 43 42 31 32 33 37

No evidence of tenderness was elicited by the patient on palpation of muscles of mastication and vestibule in all four quadrants. The patient had severe trismus (nil mouth opening). Panoramic radiograph was taken. An incidental finding revealing an ovoid homogenous radiolucent area of size 4mm in diameter was found below right inferior alveolar canal. (Fig 1) It was asymptomatic. Provisional diagnosis of Stafne cyst was given. Patient refused to undergo biopsy as patient wanted to address problem of post extraction trismus only. Patient was advised muscle relaxants and hot fomentation.

DISCUSSION

It is usually discovered by chance during routine dental radiography. Radiographically, it is a well-circumscribed, monolocular, round, radiolucent defect, 1-3 cm in size, usually between the inferior alveolar nerve and the inferior border of the posterior mandible between the molars and the angle of the jaw. It is one of the few radiolucent lesions that can occur below the inferior alveolar canal. [8] Type I (Indentation type): The inferior border of the mandible is involved. Type II (Medium type): The lesion is situated above the inferior border of the mandible but below the mandibular canal. Type III (Deviation type): Deviation of the mandibular canal is suspected. Type IV (Anterior type): The radiolucency is located in the anterior portion of the mandible. [9] The differential diagnosis includes benign and malignant jaw lesions such as odontogenic cystic lesion, non-ossifying fibroma, fibrous dysplasia, vascular malformation, focal osteoporotic bone marrow defect, brown tumor of hyperparathyroidism, ameloblastoma, basal cell nevus syndrome, giant cell tumor, or a metastasis from a primary malignant tumor. [10] Therefore, in some cases more confirmatory diagnostic tools are

mandatory. Neoplasms, such as metastatic squamous cell carcinoma to the submandibular lymph nodes or a salivary gland tumour, could create a similar appearance but rarely have such well-defined borders and can usually be palpated in the floor of the mouth or submandibular triangle of the neck as a hard mass. Although Stafne Bone Cysts are identified usually on panoramic radiographs, lateral oblique mandible projections can also be helpful for diagnosis. Lateral oblique mandible projections are commonly used to examine the body or ramus of the mandible when panoramic imaging is not available or when an image with greater resolution is needed. However, two problems associated with lateral oblique mandible projection are image distortion and superimposition of the cervical vertebrae to the site of interest. [11] Computed tomography (CT) and clinical exam is typically sufficient to distinguish between Stafne defect and above mentioned lesions. The Stafne defect also tends not to increase in size or change in radiographic appearance over time. Hence the term "static bone cyst" has been coined. Tissue biopsy is not usually indicated, but if carried out, the histopathologic appearance is usually normal salivary gland tissue. Sometimes attempted biopsy of Stafne defects reveals an empty cavity possibly because the gland was displaced at the time of biopsy or other contents such as blood vessels, fat, lymphoid or connective tissues. Defects of the anterior lingual mandible may require biopsy for correct diagnosis at this unusual location. The radiolucent defect here may be superimposed on the mandibular anterior teeth and be mistaken for an odontogenic lesion. Sometimes the defect may interrupt the contour of the inferior border of the mandible and may be palpable. Sialography is able to depict salivary tissue in the bony cavity and has been used to confirm the diagnosis. However, there were case reports of surgically proved Stafne bone cavity with negative results in sialography. [12] CT currently considered as the complementary test of choice, has the great advantage of verifying the peripheral origin of the lesion and the conservation of the lingual cortical, which are essential characteristics for discounting other pathologies such as apical or residual cysts, fibrous dysplasia and traumatic osseous cyst among others. The fact that CT is more specific to bone lesions of the jaws and much less so to soft tissue have led some authors to advocate magnetic resonance imaging (MRI) imaging as the primary diagnostic technique. CT will show a shallow defect through the medial cortex of the mandible with a corticated rim and no soft tissue abnormalities, with the exception of a portion of the submandibular gland. The border is well corticated and it will have no effect on the surrounding structures. [13] In recent

years, imaging techniques such as cone beam computed tomography (CBCT) and MRI have provided detailed information about definitive diagnosis of this entity in addition to panoramic radiographs. CBCT could be suggested as the suitable diagnostic modality for this bony configuration of the mandible because CBCT has the advantage of lower radiation dose compared to CT when suitable exposure parameters are selected. Also MRI is suggested to reveal the content of tissue extending into the bone cavity without surgical intervention. MRI is suggested for definitive diagnosis of this cystic lesion with the advantage of superior soft-tissue characterization, multiple imaging planes, different echo sequences and discrimination and determination of the content of the cavity without radiation exposure. [14] Branstetter *et al.* [15] were the first to establish a diagnosis of this entity merely on MRI imaging with no further treatment. The main advantage of MRI imaging is its superior soft tissue characterization and discrimination. The superior soft tissue contrast of MRI imaging should be adequate to make the diagnosis of Stafne Bone cyst, even without any intravenous contrast material. Its major disadvantage is the high cost and the distortion artefacts produced by dental material.

TREATMENT

No treatment is required, but neoplastic processes metastatic malignancy to the submandibular lymph nodes and/or salivary gland tumours should be ruled out. This is usually done with clinical exam and imaging. Very rarely, since the defect contains salivary gland tissue, salivary gland tumors can occur within an established defect but there is likely no difference in the risk of neoplasia in salivary gland tissue at other sites. Surgery is not necessary for the treatment of anterior or posterior Stafne bone defect. Surgical exploration or biopsy should be performed in atypical cases or other suspected lesions. Management of this defect is conservative with long-term radiographic follow-up. Atypical cases or other suspected lesions are evaluated with confirmatory three dimensional imaging methods and if necessary, biopsy should be performed for diagnosis. All of the patients in this study were notified about the lesion and scheduled for follow-up. [16]

CONCLUSION

Stafne bone defect was an incidental finding, presenting no evolutionary changes, and as such conservatory therapy based on periodic controls is indicated. Currently, complementary techniques such as CT are sufficient to establish a certain diagnosis.

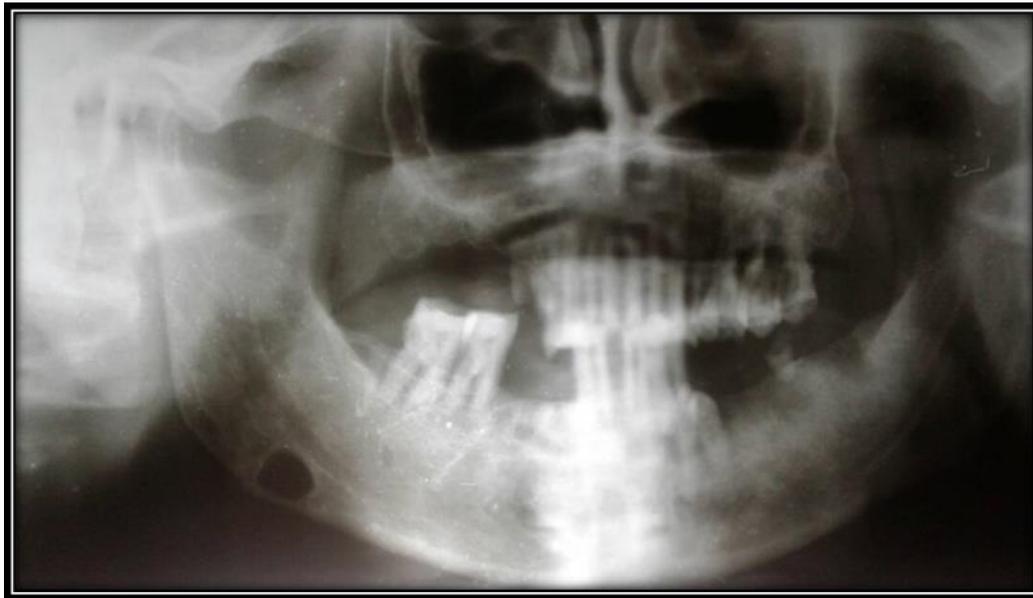


Fig 1 Panoramic radiograph showing small ovoid homogenous radiolucent area present below right inferior alveolar canal suggestive of Stafne cyst

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