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Atypical presentation of Stafne's Defect at the Mandibular Condyle: a case study from Jordan

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ABSTRACT

Stafne's defect is a rare defect of the mandible that is developmental in origin. It presents as a well-defined, corticated radiolucent lesion on the lower border of the mandible, inferior to the inferior dental canal when associated with submandibular gland. Stafne's defect above the level of the mandibular angle is rare, and as far as the authors are concerned never reported in the mandibular condyle. This case presentation reports a case of atypical Stafne's defect at the mandibular condyle in a young, medically fit, male patient, who presented complaining of pain at the left tempromandibular joint area. Diagnosis was mainly built on his Magnetic resonance imaging (MRI) and Computed tomographic Scan (CT) findings. No interventions were done regarding this lesion and patient is being followed up for 2 years. This is a case report of Stafne's defect located in an unusual anatomical place. We believe that this case report and review of the related literature will add knowledge and change clinician's way of thinking regarding these defects.

KEYWORDS: Stafne's; Mandibular condyle; parotid; idiopathic; pseudocyst.

INTRODUCTION

In the literature Stafne's defect has been referred to as: Stafne's idiopathic bone cavity, Stafne's bone cavity, Stafne's bone cyst, lingual mandibular salivary gland depression, lingual mandibular cortical defect, latent bone cyst, or static bone cyst. It is a rare defect that affects the mandibular bone [1].

In 1942, Stafne was the first clinician who described this defect thoroughly [2]. Incidence of Stafne's defect is in the range of 0.1% [3] - 0.48% [1]. Clinically, this defect affects mostly middle-aged males. It is asymptomatic and not associated with any signs and symptoms such as: swelling, erythema or discharge [1]. It is found incidentally on plain x-rays. Radiographically, this defect presents as a corticated round or oval well-defined radiolucent lesion located in the premolar area of the mandible, beneath the inferior dental canal; occasionally bilaterally [4]. The radiographic presentation of this defect is typical leading to spot diagnosis saving patients from undergoing unnecessary surgical events such as biopsy [2]. Anatomically, it is an indentation on the lingual surface of the mandibular bone and not a true bony lesion, and contains an offshoot of the submandibular salivary gland or other major salivary glands in rare cases. The latter had been confirmed by sialography showing a filling of the defect with the radiopaque media, on rare occasions, lymphoid tissue was found to be the filling material of this defect [2].

During development of the mandibular bone, entrapment of salivary glands tissues or lymphoid tissue might lead to the formation of these defects. However, after growth has ceased, these defects can occur due to subsequent lingual plate erosion made by pressure from the tissues.

Treatment is not required for Stafne's defect. Enucleation was reported as a part of the diagnostic process. Management should be limited to reassurance and follow up after radiographic diagnosis in the absence of clinical signs and symptoms, the latter may include: plain radiography, computed topography (CT) or Magnetic resonance imaging (MRI) [3]. Magnetic resonance imaging is needed in cases of unusual presentation [5].

CASE PRESENTATION

A 28 year -old medically fit male patient was referred by his general practitioner to our

oral and maxillofacial surgery clinic at the University of Jordan Hospital as a case of left condylar cyst as shown in his radiograph, presented with previously done facial CT and MRI. His major complaint was left tempromandibular joint area pain. On clinical examination, tenderness over the left joint was noted; otherwise no significant findings were noted in the head and neck.

His panoramic radiograph showed a well-defined radiolucent lesion of the left condylar neck area (Figure 1). No other abnormal findings were found. Ultrasound revealed no evidence of focal lesion. On computed tomography scans the bony defect appears to be peripheral with no soft tissue mass to support an erosive process due to a mass effect. The lesion was unifocal with no other lesion detected in the maxillofacial area (Figure 2). Final diagnosis was based on the characteristic soft tissue profile of the soft tissue residing on the peripheral bony defect in MRI imaging. The tissue was isointense to muscle and similar to the intensity of the neighboring parotid gland on T1-weighted images (Figure 3).

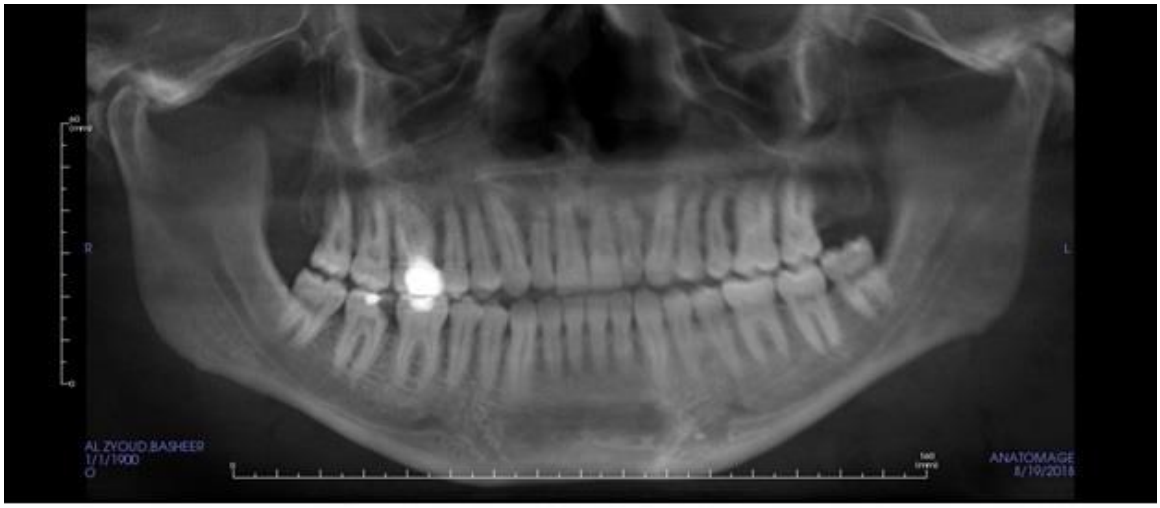

Figure (1): Reconstructed panoramic radiograph showing radiolucent lesion of the left subcondylar area

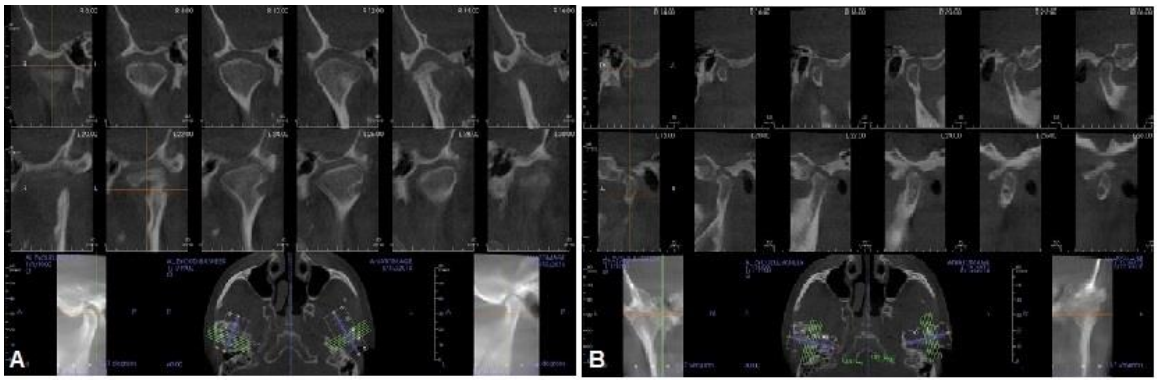

Figure (2): CBCT Scan showing a decorticated radiolucent lesion represented as depression of the left subcondylar area.

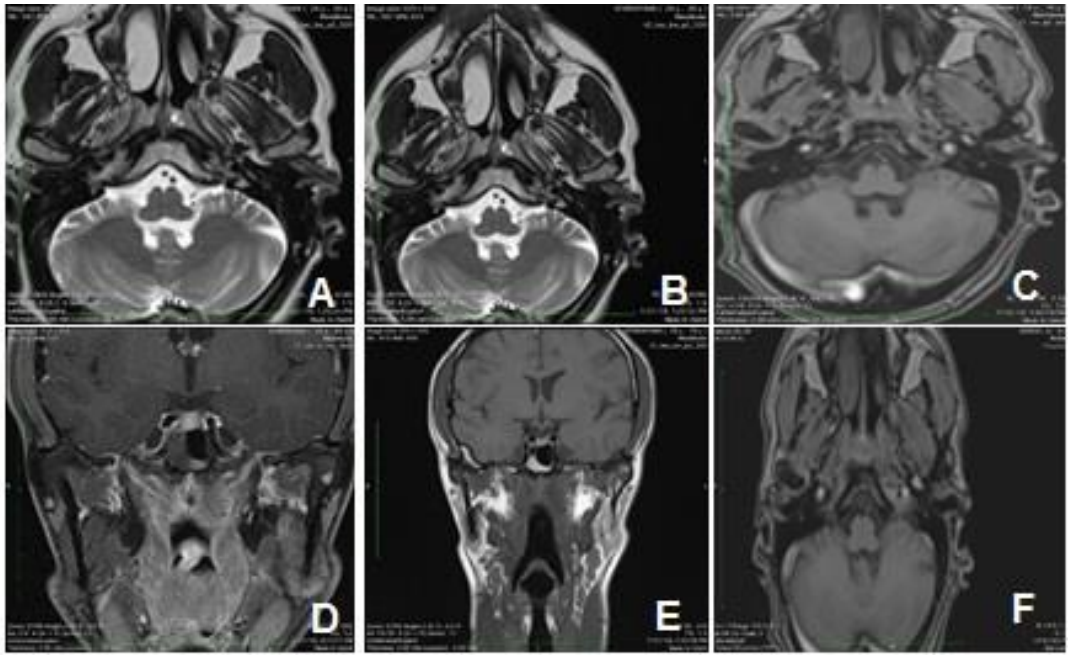

Figure (3): MRI showing salivary gland tissue similar in isdensity to the adjacent parotid gland tissue.

DISCUSSION

Stafne's defect above the level of the mandibular angle is rare, and as far as the authors are concerned never reported in the mandibular condyle. Correll *et al* looked at more than 2000 panoramic radiographs, only 13 case of Stafne's defect were detected with none of them being superior to angle of the mandible [1].

To the best of the authors' knowledge, there are only two reported cases of Stafne's defects superior to the inferior alveolar canal in the mandible. Both of the reported cases were in middle-aged males who presented complaining of pain and tenderness at the temporomandibular joint area, with no relevant medical or dental history or any other associated signs. Both lesions were detected on plain radiographs as well-defined radiolucent lesion in the ramus or condyle. Definitive diagnosis for both cases was obtained by MRI [6, 7].

The origin of these defects was debatable in the literature for decades after Stafne's description of these defects. The first assumption of this defect was congenital origin. They claim that the lesion forms due to failure of fusion in the areas previously filled by Mickel's cartilage [2]. This assumption was quickly replaced by the developmental origin hypothesis due to the following facts: the defect proximity to major salivary gland tissues, histological findings of the tissues filling the defect and absence of this defect in newborns. Pathologically, this defect is thought to originate by pressure induced by ingrowth of hypertrophic adjacent salivary gland tissues on the buccal or lingual cortex of the mandible causing bone resorption and defect formation [8].

These defects are often diagnosed in middle age male patients with mean age of 45-year-old. Presence in younger age group is rare [2]. These defects should be differentiated from other central bone lesions such as: traumatic bone cyst, benign odontogenic tumors, salivary gland diseases like pleomorphic adenoma, adjacent soft tissue lesions like lymph node pathology [9]. Buccal expansion is a rare finding with these defects, which is another clinical clue to differentiate these defects from the central lesions [10].

Diagnostic process always starts with plain radiograph .The appearance of these defects is very typical [2]. The use of computed tomography (CT) scanning and magnetic resonance imaging (MRI) is important to confirm diagnosis in the atypical lesions and to rule out the presence of centrally occurring lesion [5, 11]. Sialography had been used to detect the parotid gland extension

into the defect [12]. Wolf reported cases of ramal defects in which one case had changes suggestive of chronic sialadenitis and the other four cases had normal sialogram of the parotid gland [13].

Treatment is not required for these defects [8, 14]. However, any abnormal presentation warrants more advanced imaging modalities. These lesions are non-progressive and asymptomatic so surgical intervention unindicated in the majority of cases. However, surgical exploration and biopsy might be required to establish diagnosis especially when there are associated symptoms. In such cases, an incisional biopsy will prevent unnecessary radical treatment[15]. When surgical exploration is undertaken, salivary tissue is rarely found. However, there has been report of salivary gland inclusion in the bone cavity [12].

CONCLUSIONS

General practitioners should not be enthusiastic in their management consideration and referral criteria regarding some jaw radiolucencies that do not need any further management other than radiological modalities to confirm diagnosis. Regular follow-up with radiographic monitoring is recommended. MRI or CT scan are of paramount importance for confirming the diagnosis of Stafne's bone cavity in atypical presentations.

CONSENT

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for Editor-in-chief of this journal.

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