The unfamiliar entity in an unfamiliar location - Stafne bone cavity (Ramus variant)

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SUMMARY

Identifying Stafne bone cavities of the ramus mimics early diagnosis of cysts or tumours. In contrast, failure to recognize them can result in incorrect diagnoses and ineffective treatments. Clinical and radiological characteristics using orthopantomogram and cone beam computed tomography to make clinicians aware of such entities and help distinguish between this anatomical variant from benign tumours or cysts. This report attempts to broaden the understanding of this rare entity, as, to the best of our knowledge, only 10 cases of the ramus variant of Stafne bone cavities have been reported in the literature.

Key words: Pseudocyst – Salivary gland – Mandible – Developmental

INTRODUCTION

Preliminary radiographic evaluation of the dentomaxillofacial complex and its supporting structures is carried out by a panoramic radiograph, which has been widely accepted and clinically justified. Occasionally, this radiographic evaluation may reveal rare yet normal incidental findings. Recognition of such an entity and its characteristics in a radiographic image is detrimental to a patient's health care, the failing of which could result in an inaccurate diagnosis and treatment. One such occurrence is the Stafne bone cavity (SBC).

The Stafne bone cavity was first described by Edward Stafne (1942) as a unilateral, radiolucent, and asymptomatic cavity usually located between the lower first molar and the mandibular angle. It has been addressed by a plethora of names, including "cortical mandibular depression," "Stafne bone cyst," "Stafne bone cavity," "latent bone cyst," "aberrant salivary gland defect," "developmental bone defect of the mandible," and "idiopathic bone cavity" (Kaya et al., 2018). SBC is usually mistaken for a benign tumour or cyst because of its radiological features (Campos et al., 2004). SBCs are classified into three types: the anterior variant, the posterior variant, and the ramus variant. Only 10 cases of the ramus variant are reported in the scientific literature, which makes it a very rare entity.

In this article, we present two exceptional instances of Stafne Cavity: the ramus variant in the left sub-condylar region of the mandible was found during a routine panoramic radiographic evaluation, for which ethical clearance was

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CASE REPORT 1

A 14-year-old female patient presented with the chief complaint of forwardly placed upper front teeth. There was no history of mouth breathing, difficulty in speech, or mastication. The patient was healthy, and the medical history contained no systemic diseases or drug allergies. Intraoral examination revealed the full complement of permanent teeth except for the maxillary permanent canines, which were missing clinically. The patient was diagnosed provisionally with Angle's class I molar malocclusion with increased overjet. As a preliminary radiograph, the patient was subjected to panoramic radiography, which revealed erupting maxillary permanent canines and a well-defined homogeneous radiolucency with a thin sclerotic border measuring 1 cm in diameter in the posterior portion of the ramus, involving the left sub-condylar region (Fig. 1a). There were no perturbations to the adjacent structures, such as periosteal response or anatomical structural displacement.

The radiolucency was further investigated using Cone Beam Computed Tomography (CBCT).

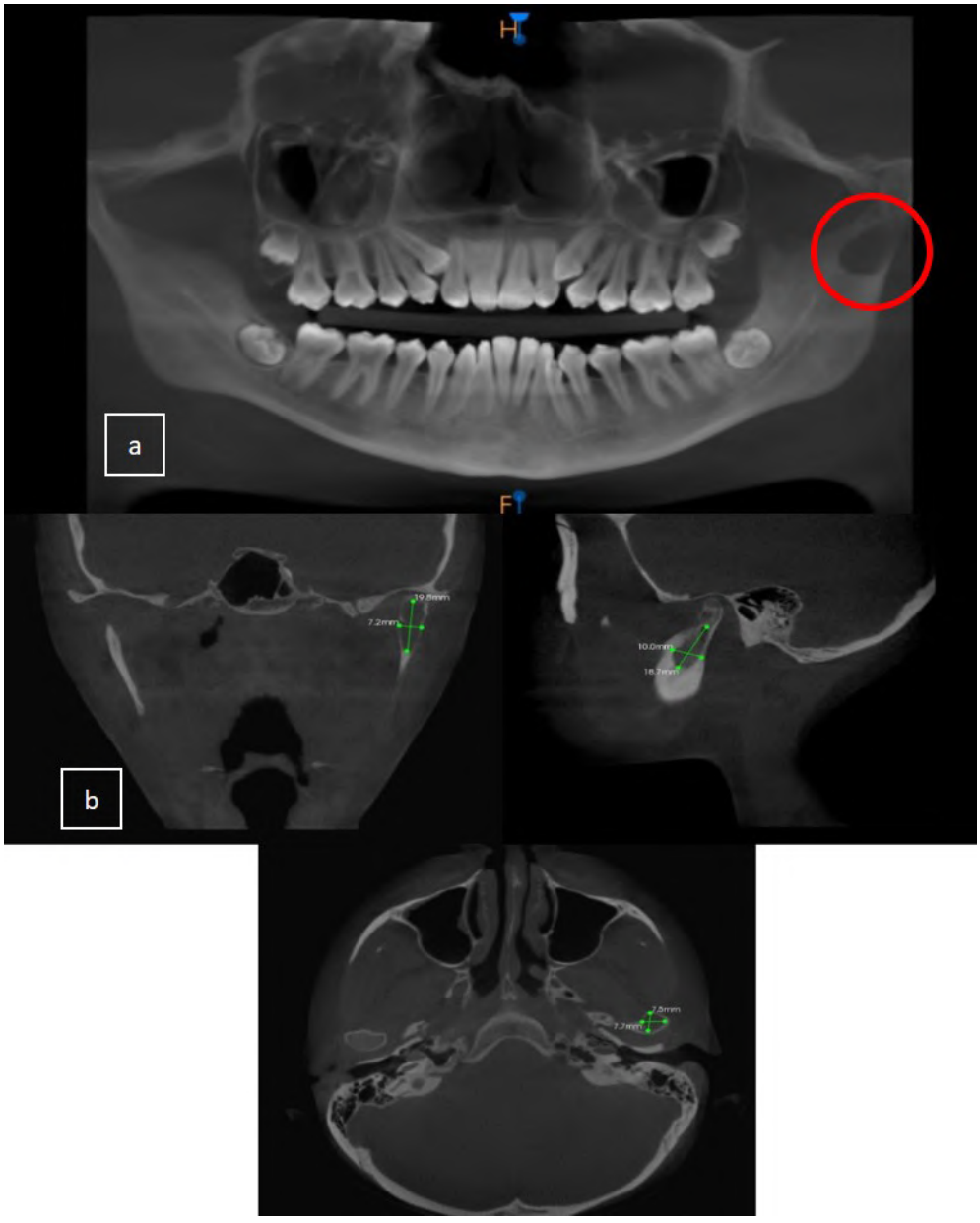

Fig. 1.- a: Orthopantomogram showing SBC of Ramus variant, noted in the left subcondylar region. **b:** CBCT showing SBC of Ramus variant.

This confirmed the uniform radiolucency with the measurement of 19.8 mm* 7.2 mm supero-inferiorly, and 7.5 mm*7.7 mm anteroposteriorly and did not signify central pathology at the left sub-condylar region. The mandibular canal was intact, and the lesion was 3 mm above the lingula (Fig. 1b).

CASE REPORT 2

Another 50-year-old male patient visited with the chief complaint of missing teeth in the right upper jaw. The patient was diabetic and was under medication. Intraoral examination revealed that the right first, second, and third permanent molars and upper right second premolar were missing. The patient had poor periodontal status and undesirable oral hygiene. A provisional diagnosis of chronic generalised periodontitis with a partially edentulous upper arch was made. A preliminary panoramic radiograph was advised as part of the periodontal treatment protocol, which depicted generalised horizontal and vertical bone loss. A well-defined, unilocular oval-shaped radiolucency was noticed on the posterior portion of the ramus below the neck of the condyle on the left side. The lesion was homogeneously radiolucent

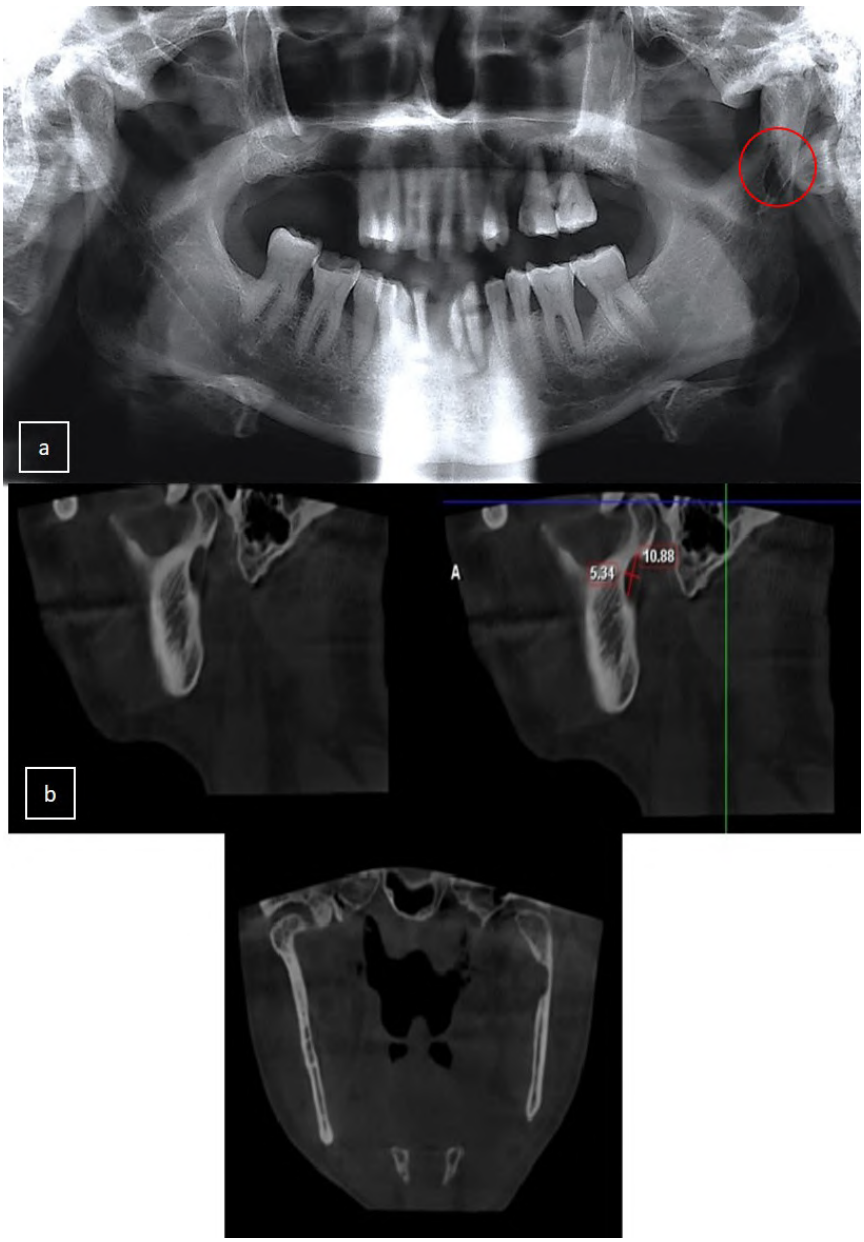

Fig. 2.- a: Orthopantomogram showing SBC of Ramus variant, noted in the posterior portion of the ramus below the neck of the condyle on the left side. **b:** CBCT showing well-defined semilunar radiolucency noted in the left medial surface of ramus below the level of subcondylar region.

without sclerotic borders and had a maximum diameter of one centimetre. There were no changes in the surrounding tissues, such as periosteal response or displacement of anatomical structures (Fig. 2a). CBCT was advised to further investigate the lesion that confirmed a well-defined, uniformly radiolucent lesion visible in the axial cross sections on the medial surface of the left ascending ramus, measuring about 10.8 x 5.3 mm with no sclerotic borders. In coronal sections, it appeared as a well-defined *semilunar radiolucency* involving the medial surface of the ramus below the level of left sub-condylar region (Fig. 2b).

DISCUSSION

A bone cavity or pseudocyst well recognized as the "Stafne bone cavity" (SBC) is primarily filled with salivary gland tissue, and may comprise of muscles, lymphoid tissue, blood vessels, fat, and/or connective tissue (Iwanaga et al., 2019). The nature of the contents of the bone cavity is determined by the local anatomical condition, as in the case of rare ramus variants, where ectopic adipose tissue development has been reported (Friedrich et al., 2012). SBC is thought to be a developmental lesion that often manifests in middle-aged to older adults, but is presumed to have its origin in intrauterine growth (Schneider et al., 2014). Stafne claimed that, during the development of the mandible, Meckel's cartilage was replaced by bone tissue and a fragment of the salivary gland was trapped. However, there are numerous hypotheses regarding its etiopathogenesis, but the most prominent one, which is commonly acknowledged by several authors, is that it occurs as a result of bone resorption caused by external pressure exerted by nearby salivary glands (Ariji et al., 1993). This hypothesis was supported by discovering large bone cavities packed with salivary gland tissue after examining 15 patients using CT scans and sialography. Certain authors, nevertheless, refuted this hypothesis by affirming the presence of some clear bone cavities with no sign of salivary gland tissue, as well as indicating similar findings on the buccal surface of the mandible, which are evidently distant from the salivary gland tissue (Friedrich et al., 2012). Clinically, it is asymptomatic, seldom palpable due to the missing bone surface, and self-limited in progression, but invasive procedures may be performed if it is incorrectly labelled as a tumour or a true cyst (Lucas et al., 2021). For instance, when it develops in an edentulous region of the mandible, the more anteriorly placed SBC variation might occasionally unintentionally resemble a remnant cyst (de Courten et al., 2002). The Stafne bone cavity (SBC) usually manifests radiologically as a well-defined, radiolucent lesion below the level of the mandibular canal in the molar-mandibular angle region.

Stafne Bone Cavities were classified into three variants according to their location (Sisman et al., 2012), as described:

- Anterior variant: The lesion present in the sublingual gland area, involving anterior region or body of the mandible.
- Posterior variant: The lesion present in the submandibular gland area, involving the posterior aspect of mandible, was the most typical site.
- Ramus variant: The lesion present in the parotid gland area, involving the ramus of the mandible.

In 1985, Wolf described the "ramus variety" of Stafne's bone cyst in the parotid area. Stafne bone cysts, which form as a result of unusual alterations in the ascending ramus, are benign, but their radiographic features may mimic some intrabony neoplasms (Bornstein et al., 2009). Furthermore, this entity should be distinguished from a pseudo foramen known as "medial sigmoid depression," which also manifests as a radiolucency found in the mandibular ramus (Langlais et al., 1983). The ascending mandibular ramus has been identified as the least prevalent site for Stafne's bone cavity, and two such cases are presented here.

Hisatomi et al. (2019) reviewed 91 instances with Stafne bone cavities, and discovered that the posterior variation was present in 0.10% to 0.48% of cases. Anterior variants are almost seven times less prevalent than posterior variants. However, in contradiction of the above statistics, we are reporting two cases with Stafne bone cavities of the ramus variant, which have been rarely reported previously in the literature (Table 1).

Table 1. Past literature reporting of Stafne Bone Cavities (Ramus variant).

Case No.	Author (year)	Age & Gender	Site	Imaging modalities performed
1.	Barker (1988)	60 year male	The posterior border of the left ascending ramus below the neck of the condyle	Panoramic
2.	Minowa et al. (2003)	63 year female	The posterior border of the right ascending ramus below the neck of the condyle	CT
3.	Minowa et al. (2003)	50 year male	Left mandibular ramus	CT, MRI
4.	Tarım Eratas et al. (2013)	55 year male	Right mandibular ramus at the junction of ramus and coronoid process	CBCT, MRI
5.	Campos et al. (2004)	14 year male	Right mandibular ramus	Panoramic, CT
6.	Anbiaee et al. (2016)	55 year male	Posterior and upper one-third of the right mandibular ascending ramus und1er the condylar neck	Panoramic, CBCT
7.	Melnichenko et al. (2016)	57 year female	Left mandibular ramus	CBCT
8.	Chen et al. (2016)	52 year male	Superior region of the left ascending ramus	Panoramic, CBCT
9.	Hisatomi et al. (2019)	52 year male	Superior posterior border of left mandibular ramus	Panoramic, MRI
10.	Lee et al. (2019)	57 year male	Left mandibular ramus	Panoramic, CBCT
11.	2023 (Current case)	14 year female	The posterior border of the left ascending ramus below the neck of the condyle	Panoramic, CBCT
		50 year male	The posterior border of the left ascending ramus below the neck of the condyle- lingual variant	Panoramic, CBCT

Although the ramus is extremely closely related to the external auditory meatus, it was challenging to palpate the jaw medially in these cases, and there was no palpable deformation of the mandibular cortex. No other symptoms were observed in either of the cases. Confirming the diagnosis of an SBC will be highly conducive when a rare or uncommon variation is suspected. Additional imaging, such as computed tomography or magnetic resonance imaging, as well as documentation of a radiolucency of unaltered size and appearance, would help to confirm the diagnosis. In a study on SBCs, Sisman et al. (2012) found that there was no discernible difference between CBCT and commonly used Multislice Spiral Computed Tomography (MSCT) in terms of accuracy. In addition, CBCT produces a lower radiation exposure dose than MSCT; therefore, it is preferred to avoid subjecting patients to radiation inadvertently. According to Katz et al. (2001), CBCT also provided detailed information about the conclusive diagnosis of SBC. As reported by More et al. (2015), an asymptomatic patient with a definitive radiographic diagnosis and periodic follow-ups are satiable, and a biopsy to confirm the diagnosis is redundant. Therefore, in the current cases, the CBCT analysis was sufficient to rule out SBC. When clinicians come across a radiolucency in panoramic radiography, Stafne's bone cavity is less often taken into consideration during differential diagnosis. However, as the lesion is asymptomatic and non-progressive, surgical intervention is unnecessary. The best long-term choice is generally considered to be periodic radiographic follow-up. The treatment of an anterior or posterior Stafne bone cavity does not require surgery. Atypical instances or any suspected lesions should be subjected to surgical examination or biopsy.

CONCLUSION

Stafne's bone cavity is a relatively uncommon anatomic anomaly that can typically be detected radiographically, although it can present a diagnostic challenge if it manifests in an unusual position. To prevent a misleading diagnosis, it must be distinguished from other cystic lesions. A surgical examination or biopsy should be performed on unusual cases or any suspected lesions. This case series attempted to further understand the occurrences of Stafne cysts, which could frequently be misdiagnosed as potentially lethal mandibular

cavities. Hence, they should not be missed out from the list of differential diagnoses, while considering cystic lesions involving the mandible.

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