Unveiling the enigma: bilateral Stafne bone defect as an incidental discovery

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SUMMARY

Diagnosing maxillofacial pathologies is challenging as they comprise various tissues, especially the jaws, due to the presence of teeth and their remnants. However, it becomes difficult when the lesion is asymptomatic, and it is critical to rule out whether there is anatomical or pathological variation. As a result, it becomes mandatory to analyze the jawbone before considering dental treatments like implants. The current case presents a rare developmental multilocular bilateral mandibular defect, marking its first publication in the literature. Higher imaging modalities assisted in diagnosing a bilateral Stafne cyst.

Key words: Bilateral – Salivary gland – Cone Beam Computed Tomography – Multilocular – Stafne bone defect

INTRODUCTION

It is clinically and generally agreed that a panoramic view is the best way to get an initial radiological look at the dento-maxillofacial complex and the parts that support it. Routine radiographic screening may, on occasion, reveal atypical but distinctive incidental abnormalities. Such an enigmatic radiographic presentation always poses questions regarding etiology, clinical significance, and an appropriate approach for diagnosis and management. The Cone Beam Computed Tomography (CBCT) imaging modality has a higher rate of identifying incidental findings of about 78.6%, out of which 14.9% persist as an osseous defect (Braun et al., 2022). Lopes et al. state that 27.55% of the incidental findings in CBCT required treatment, whereas 43.46% did not require treatment. Of the results, 28.97% required the acquisition of additional diagnostic imaging for monitoring (Lopes et al., 2016). The mandible's bilateral radiolucency is one of the rare incidental findings encountered during routine examinations.

With the help of higher imaging modalities, the appropriate diagnosis of such abnormalities may alter treatment planning, especially during surgical procedures like implant placement. This article focuses on "bilateral Stafne bone defect," which is a rare condition with only seven cases reported in the literature so far. We present a Stafne bone defect (SBD) with multilocular appearance on the left side, which is extremely rare, with only 1 case reported in the previous scientific literature.

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CASE REPORT

A 59-year-old male patient presented with a chief complaint of missing teeth in the left upper back tooth region. The patient gave a history of extraction before 1 year. Prior to 1 year, the patient received a diagnosis of hypertension and type II diabetes mellitus, underwent angioplasty, and is currently on medication. Intraoral examination revealed generalized gingival recession with deep periodontal pockets and few missing teeth, suggesting generalized periodontitis and partially edentulism of the upper arch. The periodontal therapy approach included a preliminary panoramic radiograph, which exhibited widespread horizontal and vertical bone loss. Moreover, a bilateral radiolucent lesion with a well-defined unilocular oval-shaped radiolucency on the right inferior border of the mandible and another well-defined multilocular radiolucency on the left body of the mandible were noteworthy (Fig. 1).

Cone Beam Computed Tomography was advised in order to understand the extent of the radiolu-

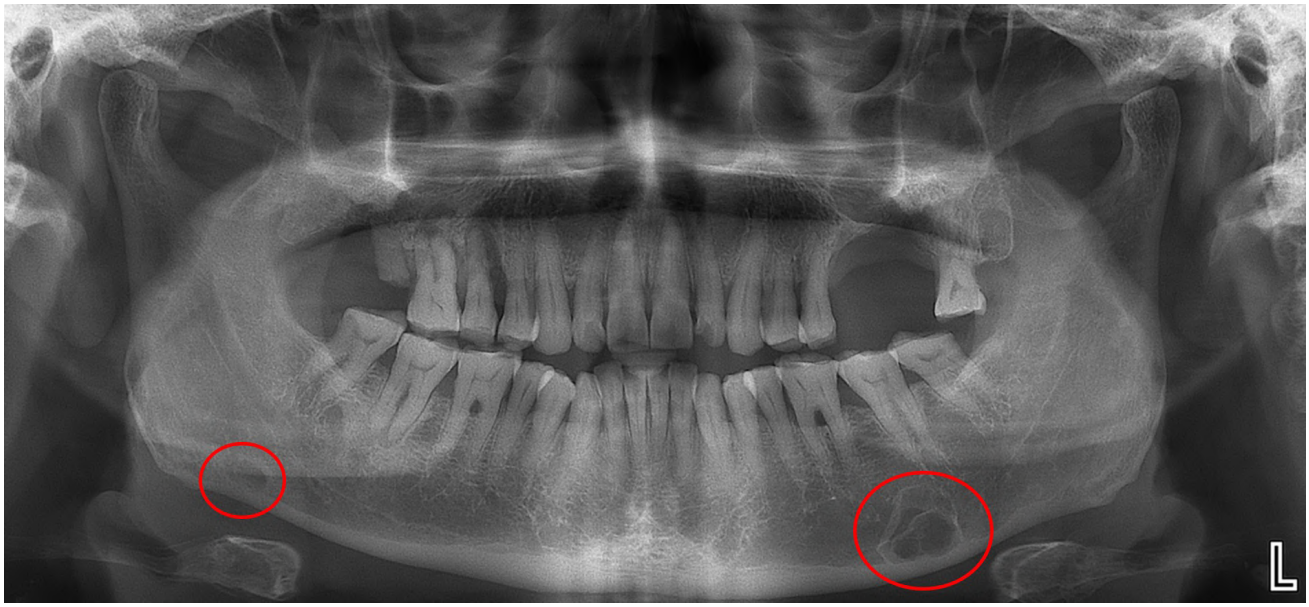

Fig. 1.- Orthopantomography representing bilateral radiolucency below the inferior alveolar canal; right side shows unilateral radiolucency, left side shows multilocular radiolucency.

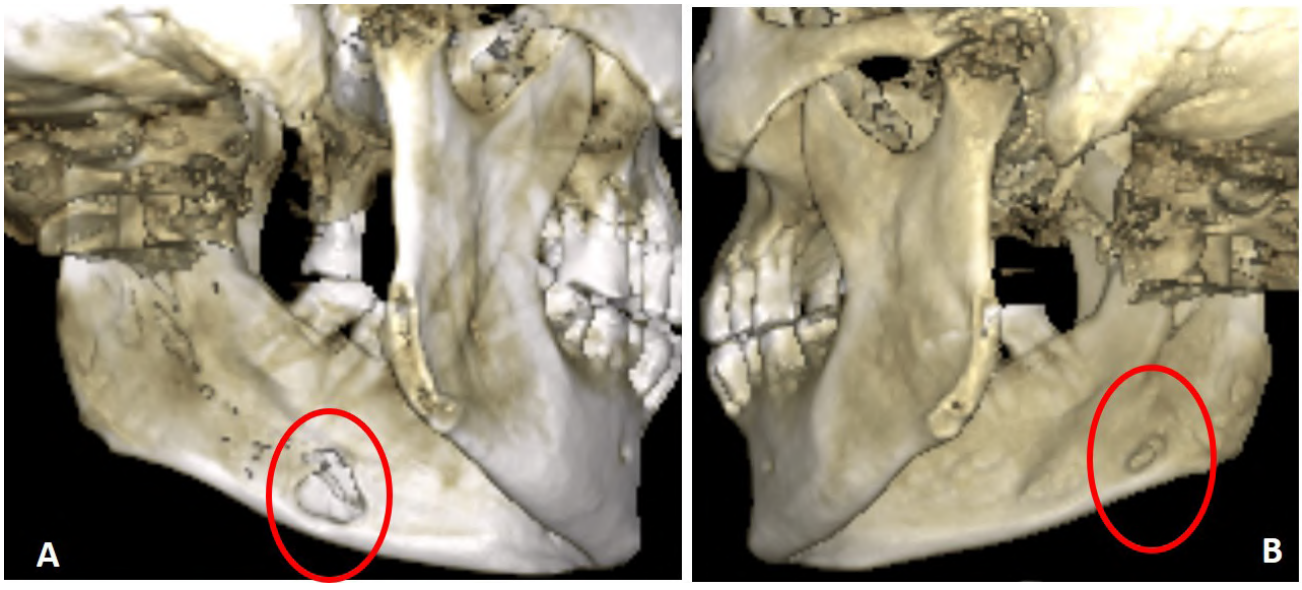

Fig. 2.- A, B: 3D Reconstruction using volume rendering that displays the bilateral concavities on the lingual cortical surfaces.

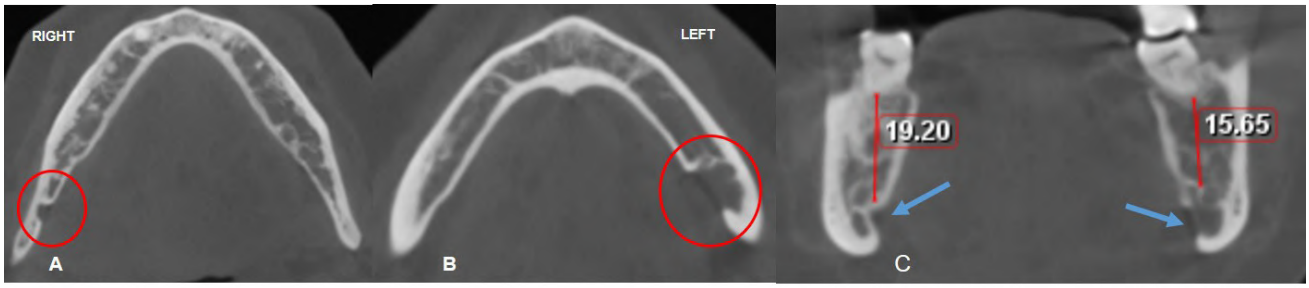

Fig. 3.- A, B: Axial section of CBCT showing bilateral radiolucency involving the lingual cortical plate of mandible. **C:** Coronal section of CBCT showing distance from the alveolar crestal margin till the semilunar radiolucency of the lingual cortical plate.

cency, which has been illustrated in the figure along with its dimensions (Figs. 2 and 3). The distance from the alveolar crest to the bilateral radiolucency on the coronal section is 19.20 mm on the right side and 15.65 mm on the left side (Fig. 3C). It was finally diagnosed as a bilateral SBD with multilocular presentation on the left side. The lesion was disclosed to the patient and advised for scintigraphy, but the patient was not willing since it is an asymptomatic condition. Therefore, upon recalling the patient for follow-up after six months, the panoramic image showed no apparent alteration.

DISCUSSION

The defect is known by a variety of terms, including cortical mandibular depression, Stafne bone cyst, Stafne bone cavity, latent bone cyst, aberrant salivary gland defect, developmental bone defect of the mandible, idiopathic bone cavity, lingual bone defect, lingual mandibular bone depression, developmental salivary gland defect, and lingual posterior mandibular bone concavity (Kaya et al., 2018). The exact cause of "SBD" is still unknown, but the most popular theory is a "glandular" one. This theory says that the submandibular gland presses down on the lingual surface of the jaw, which causes the lingual cortical plate to break down and the mandible to lose its lingual function (Schneider et al., 2014). Stafne (1942) asserts that during mandibular development, Meckel's cartilage replaced a portion of the salivary gland as bone tissue. Despite the existence of numerous hypotheses, Lello's widely accepted one proposes that relative hypoxia, a condition where the facial artery compresses a region near the mandibular lingual cortex, causes an inadequate blood supply and reduced nourishment, is the cause of the impairment (Lello et al., 1985). Salivary gland tissue primarily makes up the SBD, but it can also include blood vessels, muscles, lymphoid tissue, fat, and/or connective tissue (Iwanaga et al., 2019).

Three distinct types of SBDs are recognized radiographically, based on their location: the posterior variant, the anterior variant, and the ramus variant. The anterior variation of SBD is extremely uncommon, with a frequency of 0.009-0.03%, whereas the posterior variant is more common, ranging from 0.10-0.48% (Strom et al., 1987; Langlais et al., 1976). In contrast to our situation, a 2013 case study (Erta's et al., 2013) documented two instances of SBD localized to the right ramus. Interestingly, the literature likely first documented the extremely uncommon triple SBD (Luciana et al., 2011). The bilateral presentation of the posterior variant has been identified as the least prevalent site for SBD, which has been documented here. SBDs generally appear unilaterally in the available research, although there have been occasional instances of bilateral occurrences (Aguiar et al., 2011). Though multilocular occurrence and double unilateral occurrence are uncommon (Aguiar et al., 2011, Campos et al., 2004), in our case, the defect manifests as multilocular on the left side.

Because of the patient's lack of aberrant symptoms, the real rate may be higher than documented in various reports (Sisman et al., 2012). It is rare; however, it can occasionally result in jaw expansion. Minowa et al. and Smith et al. conducted retrospective studies, revealing that the fifth and sixth decades were the most affected, which is in accordance with our report (Minowa et al., 2003; Smith et al., 2007). In a meta-analysis of 355,890 individuals, Phanthip et al. shortlisted the SBD of

bilateral presentation according to site, size, and location (Table 1) (Phanthip et al., 2023).

The differential radiographic diagnosis of SBC includes metabolic disorders like mucopolysaccharides, multiple myeloma, multiple odontogenic keratocysts, bilateral dentigerous cysts, traumatic bone cysts, and focal osteoporotic bone marrow defects (Bayrak et al., 2020). Liang et al. say that the cysts mentioned above are always found above the inferior alveolar canal. On the other hand, SBD is usually found at the mandibular angle below the inferior alveolar nerve canal and has a specific look that helps doctors make the right diagnosis (Liang et al., 2019). Similarly, our case shows that the bone defect visible on CBCT is located lingually, beneath the inferior alveolar canal, which helps rule out other potential radiological differential diagnoses.

In some cases, more imaging tests, like magnetic resonance imaging, contrast-enhanced computed tomography, salivary gland scintigraphy, or positron emission tomography, are needed to make a diagnosis. These tests would rule out the metabolic activity of tumor cells. Another conventional diagnostic technique, sialography, identifies the salivary gland as the origin, but its 2D planar imaging limits its use. Nonetheless, patients find this technique to be intrusive and painful (Dobrin et al., 1988).

Because SBD is a benign condition that does not result in pathological alterations, surgical intervention is no longer necessary. However, in the present era of various types of dental implants, the recognition of such defects preoperatively becomes extremely important to avoid post-operative complications. Conversely, for larger defects, a thorough and definitive radiological investigation may indicate the need for bone trephining and bone exploration. An iliac bone transplant may be an option while planning for prosthetic rehabilitation following a cystectomy with a den-

Table 1. Past literature reporting of Stafne bone cavities (bilateral posterior variant). IOPAR- Intraoral periapical radiograph, CT- Computed Tomography, CBCT- Cone Beam Computed Tomography.

CASE NO.	AUTHOR & YEAR	AGE & GENDER	LOCATION	IMAGING MODALITIES
1.	Boyle et al. 2000	57 Year Male	Bilateral body of mandible & left parasymphysis region	IOPAR, Panoramic, CT
2.	John R. Lukacs et al. 2002	Dry specimen Male Dry specimen Female	Bilateral defect of anterior mandible Bilateral inferior to right & left third molars	- -
3.	Lelia M. Guedes Queiroz, 2004	32 Year Female	Bilateral defect of anterior mandible	IOPAR, Panoramic
4.	Elif Tarim Ertas, 2013	55 Year Male	Bilateral- Lingual ramus & Buccal ramus	CBCT, MRI
5.	Carlos et al. 2013	37 Year Female 73 Year Male	Bilateral body of mandible Bilateral angle of mandible	Panoramic Panoramic
6.	Alaettin et al. 2019	58 Year Male	Bilateral body of mandible	Panoramic
7.	Mayank, 2023	62 Year Male	Bilateral body of mandible	Panoramic, CBCT
8.	Present case, 2024	59 Year Male	Bilateral body of mandible	Panoramic, CBCT

tal implant (Alzahrani et al., 2023). In the present case, follow-up was done in six months, and no other changes were noted. However, frequent follow-ups are advised to ascertain the radiographic variation.

CONCLUSION

Dental practitioners should be aware of preliminary radiographs, as they can reveal various pathological conditions that may necessitate further investigation or treatment. As clinicians encounter this asymptomatic bilateral mandibular radiolucency in patients, it becomes imperative to explore the underlying factors contributing to its existence and the necessary steps for accurate diagnosis and appropriate management, which might be destructive to the patient's health during implant placement. Therefore, when evaluating lesions affecting the mandible, one should not exclude the SBD from the list of differential diagnoses.

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