Stafne Bone Defect: A 24- Year Case Report of a Benign Surgical Condition

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Abstract

BACKGROUND: Stafne's cavity, also known as Stafne's bone defect, is a focal osseous cavity defect of the cortical tissue on the lingual surface of the mandible. It is often identified incidentally during radiographic examinations in the field of medical-dental and oro-maxillofacial care. The lesion is typically characterized by a radiolucent, oval or round-shaped area in the region between the angle of the mandible and the lower margin of the mandible, often adjacent to the lower first molar.
MATERIALS AND METHODS: This case report aims to describe the anatomo-topographical, clinical, and radiological characteristics of Stafne's defect, which was incidentally found during a routine assessment of an 80-year-old patient at the Department of Dentistry at the IRRCS San Raffaele Hospital in Milan. A unique feature of this clinical case was the ability to compile a clinical and radiological history of the patient, following an initial diagnostic assessment of the lesion approximately 24 years ago, with a historical series of radiographs and radiographic reports.
RESULTS: With access to a chronological series of anatomical radiographs, used for monitoring the evolution of the osteolytic lesion over 24 years, in line with the progression, resolution, or stability of the condition, it was possible to confirm the incidental and presumptive diagnosis of Stafne's cavity. Panoramic radiographic images, both historical and current, initially provided an assessment of the lesion. Subsequent evaluations with cone-beam computed tomography (CBCT) allowed for a more precise diagnosis and confirmed the initial clinical suspicion.
CONCLUSION: Although it is a rare osseous defect, Stafne's cavity should always be considered in the differential diagnosis of mandibular osseous lesions with a pseudocystic phenotype located both more cranially and more caudally to the mylohyoid line. The diagnosis is often incidental and tends to affect males. This condition, considered rare, is benign and does not evoke any symptoms

Keywords: Stafne's Bone defect, Benign Condition, 24-Years Follow Up, Literature Review, Benign surgical condition

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Introduction

Stafne's cavity, also known as Stafne's defect, is a focal osseous cavitary defect of the lingual cortical tissue of the mandible, often incidentally identified during radiographic investigations in the medical-dental and oro-maxillofacial fields (1,2,3,33). This morphological alteration of the mandible, exhibiting a pseudocystic phenotype, was first described by Edward Stafne in 1942 (3). Typically, it is characterized by a radiolucent oval or round-shaped area in the region between the angle of the mandible and the lower first molar, usually located caudally to the lower margin of the mandible, in the segment between this point and the inferior border of the mandible (2,4,32,39). However, there have been reported cases of anterior localization as well (27). The pathogenesis of the defect is not entirely clear, with the salivary gland hypothesis being the most widely accepted (1,5). It is believed that these cavities, often referred to as "salivary gland anatomical imprints," are attributed to pressure, proliferation, or translocation of salivary tissue or other adjacent soft tissue to the lingual cortical surface of the mandible (9). A multifactorial etiology is not excluded. The lesion can be considered an anatomical developmental defect (1,7). This osseous depression is benign, typically asymptomatic, static in both morphological and dimensional evolution, and does not require surgical treatment (5). The terminology associated with the lesion's definition is rather diverse. According to the anatomical-pathological nomenclature proposed by Neville et al. (1), Stafne's cavity is also known as Stafne's bone cyst, lingual mandibular salivary gland depression, latent bone cyst, static bone cyst, or cortico-lingual mandibular defect (1,6,15). The incidence of Stafne's cavity varies between 0.10% and 0.48% of the general population and is more commonly found in males. Patients are typically diagnosed within the demographic range of 50-70 years (11,13). A study by Chaweeborisuit P. et al. demonstrated a prevalence of the lesion around 0.17%, primarily localized at the mandibular angle unilaterally, with a higher frequency in men than women (26). Despite being a rare condition, a correct differential diagnosis allows for recognition of this condition and its exclusion from numerous other odontogenic or non-odontogenic pathologies that may present with the same anatomical location or morphological phenotype (1,7). The scientific literature has reported both unilateral and, more rarely, bilateral lesions (11, 36). The most common topographic variant is the posterior-lingual variant, located in the submandibular anatomical region (13). An anterior-lingual variant in the canine-premolar region in the sublingual region has also been described (8,19, 34, 35). With convincing pathophysiological evidence, the radiolucent defect could originate from the erosion or direct resorption of the cortical tissue that is focally involved, resulting in a peculiar osseous depression (1,7, 31). In the literature, there have also been cases of Stafne's cavity with complications related to possible apical infections of adjacent dental elements (30). Although the lesion is asymptomatic, symptomatic involvement may occur due to factors external to the lesion itself. Diagnosis of the lesion relies on the use of computed tomography (1,40), magnetic resonance imaging, and sialography (1,9,14). This clinical case report aims to describe the anatomo-topographical, clinical, and radiographic characteristics of Stafne's cavity, as encountered in a patient visiting the dentistry unit of the IRCCS San Raffaele Hospital in Milan. A unique feature of this clinical case was the ability to compile a clinical and radiological history of the patient following an initial diagnostic assessment of the lesion approximately 24 years ago. This long-term follow-up provided valuable insights into the static nature of the analyzed condition. The article also includes a brief review of the scientific-descriptive literature regarding mandibular osseous defects related to salivary tissue. The goal of the article is to emphasize the importance of differential characteristics of Stafne's depression compared to other odontogenic and non-odontogenic pathological categories, particularly those with a pseudocystic phenotype.

Case presentation - Materials and Methods

An 80-year-old male patient presented to the dentistry unit at IRCCS San Raffaele Hospital seeking specialized consultation for an acute pericoronitis episode related to tooth 38. The patient reported being referred to our department by his general dentist, who had already noted previous and recurring episodes of pain and inflammation in the affected area. As part of the initial clinical protocol upon admission to the hospital unit, the following data were collected: age, gender, past medical history, current medical history, reason for seeking dental care, any clinical documentation provided by the patient, and any radiographs taken by the patient at other radiology centers, with or without specialist radiologist reports.
A preliminary dental examination was conducted to investigate, confirm, or refute the reason for the patient's visit and to determine the appropriate clinical management, following a discussion with the patient. The patient had a history of mild right renal insufficiency, with no significant pharmacological history. The patient possessed radiographic documentation in the form of a two-dimensional radiograph printed on a transparent sheet, corresponding to an orthopantomographic study of the dental arches, which was dated. The associated radiologist's report described a "fibro-dystrophic depression" at the right mandibular angle. In addition, the patient had four additional radiographic transparencies showing an axial tomographic study of the lower dental arch, conducted using spiral technique, which provided anatomical reproductions in axial planes and reconstructions in frontal and para-axial planes, in actual size (1:1 scale). This examination included reconstructions of the right hemi-mandible, where a radio-transparent osteolytic area was identified between the mandibular angle and the lower right third molar, just below the mandibular canal. According to the axial planes, images of the region were also studied with an algorithm for the assessment of soft tissues. The radiologist's report, attached to the patient's tomographic documentation, described a focal osseous concavity and specified that the radiographic image characteristics were consistent with those of a "salivary gland developmental fossa" on the lingual surface of the mandible. The radiologist further noted that the borders of the depression, particularly deep, appeared sharp and regular. Additionally, it was mentioned that the salivary gland was adjacent to the observed osseous cavity, without extending into its lumen, which was in line with common findings in congenital osseous defects of this nature. The specific term "Stafne's cavity" had never been attributed to the developmental fossa in the radiology report.

Considering the dating of the patient's radiographic examinations, a decision was made to perform a current diagnostic assessment, taking into account the patient's current clinical situation, as it was not possible to obtain this information from examinations conducted more than 20 years ago. The execution of this examination, primarily aimed at defining the clinical picture of acute pericoronitis of tooth 38 (which remained the primary indication for the examination), was therefore justified.

A cone-beam computed tomography (CBCT) three-dimensional investigation was performed to study the anatomical relationships of tooth 38 with the vestibulo-lingual boundaries of the mandibular body and the mandibular canal. This study, conducted exactly 24 years after the previous one, also revealed a lesion that did not differ in its morphological and topographic characteristics from a radiolucent mandibular defect for which the patient had previously received a presumptive diagnosis during the previous investigations ( ).

Furthermore, following classical semiotics principles, the technique of intra and extra-oral inspection and palpation was employed. No swelling or continuity solution was identified in the investigated anatomical region. Bimanual palpation of the oral floor with opposing fingers yielded negative results. While running a finger along the postero-medial face of the mandible beneath the adherent gums and along the cortical wall adjacent to the tongue, no depressions or discontinuities of the lingual osseous plate could be appreciated by touch. However, it should be noted that the soft tissues overlaying the bony walls often make direct clinical evaluation challenging, especially in the area corresponding to the oral floor. Based on patient history and palpation, local-regional symptoms were absent. Sialography was not performed to reasonably determine glandular filling of the defect, nor was magnetic resonance imaging conducted.

Radiographic follow-up

Having access to a chronological series of anatomical radiographs, spanning a 24-year period and aimed at monitoring the evolution of the osteolytic lesion, in accordance with the possible progression, resolution, or stability over time, it has become evident that the incidental and presumptive diagnosis of Stafne's cavity can reasonably be confirmed. Orthopantomographic images, both remote and current, provided an initial evaluation of the lesion. Special attention was paid to the apical area of the canine-premolar region to exclude an anterior-lingual variant of the defect. In this region, no notable signs were observed. The two-dimensional images revealed a unilateral, well-defined, radiolucent, unilocular, and partially bilobed lesion caudally. It was located between the posterior end of the right mandibular body, in the region of the lower right third molar, and the angle of the mandible, below the inferior alveolar canal. The anatomical variant was posterior-lingual, and the localization was typical. The peripheral border appeared osteosclerotic, and the morphological phenotype was pseudocystic. The focal osseous defect did not interrupt the continuity of the lower margin of the mandible and bordered superiorly on the caudal limit of the mandibular canal. The apices of the roots of the elements immediately cranial to the osseous cavity were radiographically free of pathology and did not exhibit peri-apical osteolytic areas or resorption areas. There was no history of maxillofacial trauma in this region. Examination revealed edentulism of tooth 47, which was not in topographic contiguity with the osseous depression.

Both remote and current CT images revealed the interruption of the continuity of the lingual cortical plate of the right hemi-mandible in the focal osteolytic area. The buccal cortical plate at this point was rather thin. The

Figure 1. Orthopantomography (1998) of the dental arches showing a radiolucent lesion on the right mandible that is similarly consistent with a diagnosis of Stafne's Cavity.

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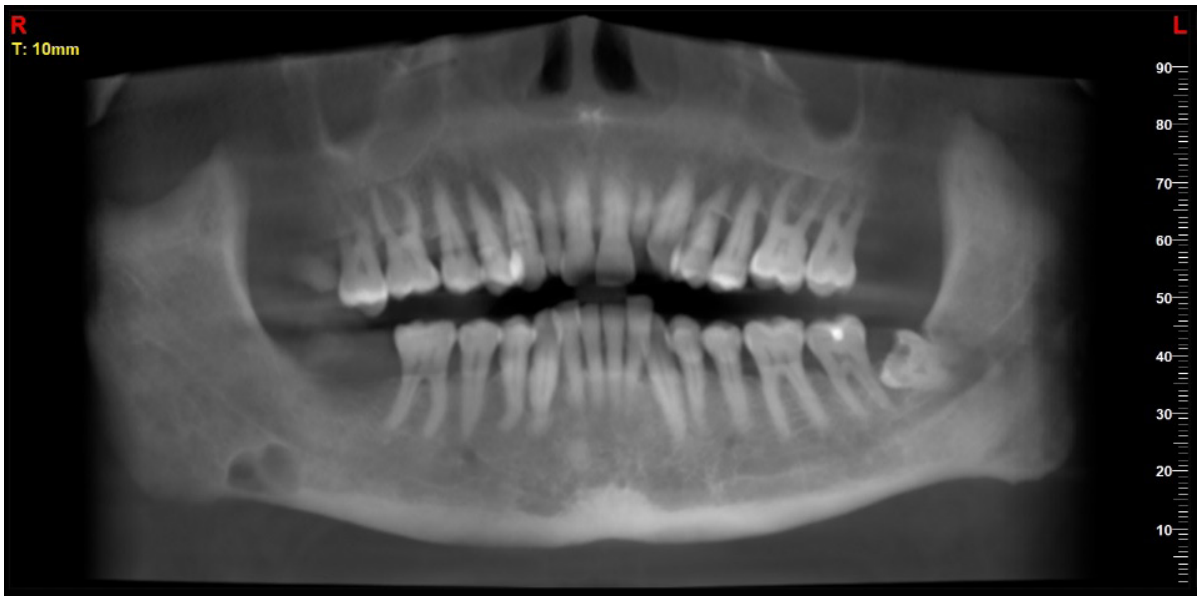

Figure 2. CBCT-like panoramic view (2022) shows the radiolucent lesion on the right mandible, which is consistent with a diagnosis of Stafne's Cavity. Compared to the radiograph from 1998, the lesion does not appear to have changed over time.

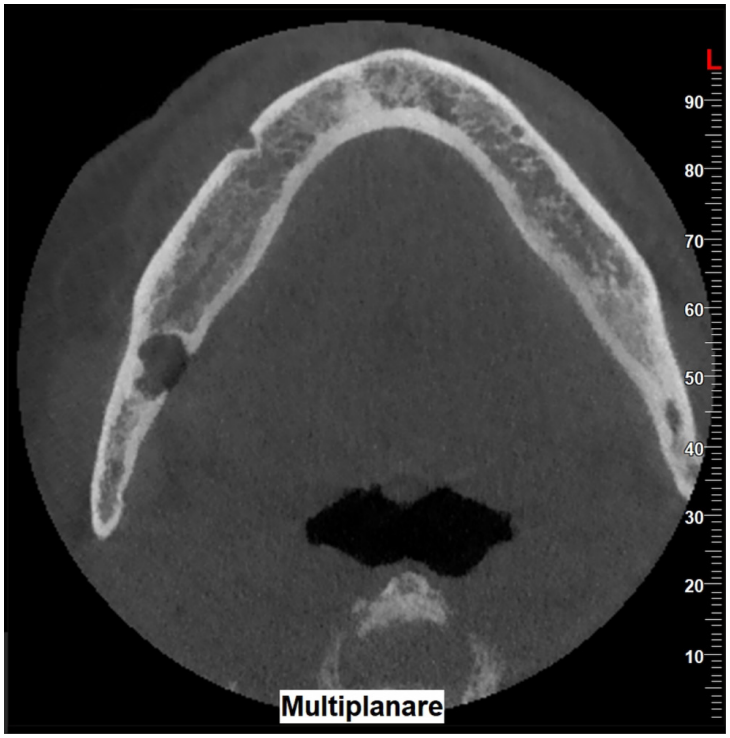

Figure 3. CBCT coronal view (2022), in this image, the outline of the cavity is clearly visible along with a reduction in the buccal cortical bone.

osseous concavity was focal. In the coronal plane, the defect extended lingually-vestibularly, assuming a bi-lobed morphology. The two lobes or "salivary imprints," slightly outlined, were oriented vestibularly. The major postero-anterior diameter was measured at 12.77 mm. In the para-axial cross-sectional reconstruction, the depression was located just caudal to the lower limit of the mandibular canal in the region of the lower third molar, with continuous and slightly reniform coronal margins.

From the radiodiagnostic follow-up, it is evident that the focal osteolysis is represented by a thinning of the mandibular cortical on the buccal side (antero-medial wall of the mandibular body, posterior region) and a depletion of lingual cortical bone tissue in the discontinuity (postero-medial wall, posterior region). Radiographic comparison, performed by comparing coronal and sagittal anatomical sections of the radiographic series in our possession, did not show any change in the dimensions of the lesion. This observation implies that the osseous defect has presumably remained stable over time, in terms of size and morphology, consistent with the static behavior of the cavity.

Due to the absence of lesion atypia or suspicion of non-benign pathology, surgical exploration or biopsy was not deemed necessary. The review by Soares et al. (14) raises an interesting question regarding the male

predilection for Stafne's cavity. According to the authors, degenerative vascular alterations are more commonly found in middle-aged males. However, the patient's medical history does not report any such information.

Discussion

Stafne's cavity, often described as Stafne's bone cavity or defect, is a focal osteo-cavitary lesion of the mandibular body (3), typically identified incidentally during radiographic examinations of the maxillofacial region or diagnosed through radiographic examinations in the field of dentistry (1,2,28). It was first described by Edward Stafne in 1942 (3). Stafne described 35 asymptomatic mandibular bone cavities located unilaterally in the posterior region of the mandible. These defects, all radiolucent, were oval and round, always more cranial than the lower margin of the mandible and more caudal than the inferior alveolar canal (3,6). Stafne's cavity is described by some authors as mandibular lingual osseous cavity, idiopathic bone defect, benign mandibular concavity (1,7). It is also known as a static bone cyst since it typically remains stable over time, both morphologically and dimensionally (1,6). In the systematic review by Soares et al. (7), Stafne's bone defect is described as an anatomical imprint of mandibular development resulting from pressure, proliferation, or translocation of soft tissue structures immediately adjacent to the lingual cortical plate of the mandibular body, such as salivary glands or other tissues. Most cases in the literature suggest a glandular etiology (29). If pressure is exerted by the submandibular gland, the mandibular bone defect is attributed to the "postero-lingual variant" (5). The original description of "Stafne's cavity" originally referred to this anatomical location, specifically the segment between the mandibular angle and the permanent first molar, increasingly caudal to the inferior alveolar canal (1,7). If pressure is exerted by the sublingual gland, the mandibular bone defect is attributed to the "antero-lingual variant," which is less frequent but noteworthy (5). In this case, the anatomical location involves the bony segment between the areas of the lower incisors and premolars, more cranial than the tendon insertion of the mylohyoid muscle (7).

With sufficient physiological convincing, the radiolucent defect may originate from direct erosion or resorption of the cortical tissue that is focally involved, resulting in a bone depression with unique characteristics (1,7). The radiolucent area does not appear to be truly congenital, meaning it is not always present at birth. According to Neville et al. (1), most cases with this diagnosis involve non-young individuals, with childhood age rarely affected. This implies that the lesion may develop at a more advanced stage of mandibular ontogenesis or at a later stage of an individual's life as a multifactorial modification of the lingual cortical of the mandibular body. Neville et al. (1) exclude that the lesion could constitute a congenital pathology because, in rare cases, an evolution in the extent of the depression over time has been observed. It is not excluded that such mandibular cortico-lingual defects are the result of insufficient focal osteogenesis in an area previously occupied by Meckel's cartilage. Another hypothesis is that the bone depression is the result of ischemic-necrotic events or osteo-resorptive changes originating from improper pressure or facial artery perfusion (13). According to Lello et al. (5), both superior and medial vector traction muscle forces and inherent hemodynamic forces acting on the facial artery would increase the relative distance between the lingual-mandibular wall and the artery itself, resulting in decreased local-regional blood perfusion.

Clinical reports suggest that even rarer variants of the cavity may be observed. If the development of the lesion involves the parotid gland, the defect can affect the ascending branch of the mandible, both in the lingual and buccal variants (13). The bone cavity is almost always asymptomatic. Diagnosis often occurs incidentally during routine radiographic examinations or for other clinical reasons, often of a dental nature (2,4). Epidemiology is not yet fully defined, although several studies, including Sisman et al. (2), have estimated an incidence ranging from 0.10% to 0.48% in the general population. This benign osseous defect is more frequently observed in male patients, especially diagnosed between the ages of 50 and 70 (13). Radiographically, the defect appears as a well-defined, unilateral, radiolucent area, more cranial than the lower border of the mandible and more caudal than the inferior alveolar nerve (4). This characteristic is pathognomonic, and biopsy for anatomopathological confirmation is often unnecessary (16). The radiolucent area in the mandibular skeleton can be filled by an accessory lobe or extension of the submandibular or sublingual salivary gland (5). Diagnostic confirmation is provided by sialography, which confirms the presence or absence of salivary parenchyma in the bone cavities (10, 30). It cannot be ruled out that, considering cases reported in the literature, the defects are filled with lymphoid tissue, muscle tissue, fibrous connective tissue, adipose tissue, blood vessels. The bone cavity can also be patent (17). Radiolucency is often unilateral and unilocular, with a sclerotic margin. The phenotype is pseudocystic (15). Cases of bilateral and bilobed cavities are reported (13).

This type of benign pathology does not affect the need for implant rehabilitation. Current rehabilitation techniques involve the use of inclined implants (20) with techniques that minimize peri-implant bone loss (23). It is essential to ensure that implants are properly maintained over time to prevent failure, especially in elderly patients (22, 25). The differential diagnosis of Stafne's cavity is of paramount importance. Several other osseous lesions with a pseudocystic phenotype may have radiographic similarities to this depression. According to Soares et al. (7), these include odontogenic and non-odontogenic radiolucencies: periapical cysts, traumatic bone cysts, odontogenic keratocysts, dentigerous cysts, fibrous dysplasia, ameloblastoma, and focal osteoporotic marrow defect. The diagnosis should also exclude fibro-osseous lesions, traumatic bone cysts, eosinophilic granulomas, multiple myeloma, and metastatic mandibular osteolysis. For most cases, periodic monitoring with radiographic examinations may be sufficient to track any changes in the lesion over time. However, in the presence of doubtful cases or anatomical changes in follow-up radiographic images, more sophisticated diagnostic techniques may be necessary, combining computed tomography with magnetic resonance imaging (12,14,18, 37,38). The main advantage of magnetic resonance imaging (MRI) is its superior ability to characterize and discriminate soft tissues (14,18). Treatment, if needed, should be decided on a case-by-case basis and should not be confused with therapeutic options for other oral bone lesions (24). Surgery is not necessary

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for the treatment of the bone defect (10). Surgical exploration or biopsy should only be performed in atypical cases or in the presence of suspicious lesions. In conclusion, regarding the presented clinical case, it can be noted that the diagnosis of Stafne's lesion at the San Raffaele Hospital Dentistry Unit in Milan was incidental, as often reported in the literature. The patient did not exhibit any symptoms or pathological signs associated with the observed osteolytic depression. The patient came to our observation for a dental reason unrelated to the anatomical region under study here. In exploring the etiological theories behind the formation of Stafne's cavity, our analysis highlights the 'glandular pressure' hypothesis as a compelling explanation (1). This viewpoint suggests that the cavity arises from pressure exerted by adjacent salivary glands on the mandibular bone during its development, leading to localized erosion or thinning of the bone cortex, thereby forming Stafne's cavity. The stability of these cavities over time and their proximity to salivary glands underscore a developmental, rather than pathological, origin. Our advocacy for the glandular pressure theory stems from its consistency with the clinical and radiographic features observed in Stafne's cavities, such as their specific anatomical positioning, morphological stability, and the presence of glandular tissue within some cavities. This hypothesis provides a clear explanation for the developmental onset of the cavity and supports the non-progressive nature of this anatomical peculiarity. The glandular pressure hypothesis emerges as a logical framework for understanding the formation and enduring presence of Stafne's cavity, resonating with both the empirical observations and the conservative management these lesions usually require.

Conclusion

Although a rare bone defect, it's imperative that Stafne's bone cavity (1942) is consistently included in the differential diagnosis of mandibular bone lesions with a pseudocystic appearance, whether located more cranially or caudally relative to the mylohyoid line. The diagnosis is often incidental and exhibits a predilection for males. As elucidated in the discussion, this benign osteo-cavitary anomaly presents itself typically in individuals aged between 50 and 70, and its discovery often occurs during routine radiographic examinations or for other dental-related reasons. Only a precise nosological understanding of this pathological entity, confirmed through CT and/or MRI analysis, can exempt the clinician from implementing inappropriate surgical and dental procedures. The literature suggests a glandular etiology for most cases of Stafne's cavity, with variations based on the adjacent soft tissue structures exerting pressure on the mandibular body. These variations, namely the postero-lingual and antero-lingual variants, underline the importance of a meticulous radiographic analysis in discerning the exact nature and etiology of the bone defect. Treatment of the static posterior-lingual lesion is not generally necessitated, emphasizing a benign prognosis. Furthermore, the systematic review by Soares et al. sheds light on the potential mechanisms underpinning the development of this lesion, be it due to direct erosion or resorption of the cortical tissue or other multifactorial modifications of the lingual cortical of the mandibular body at a later stage of an individual's life. These insights reinforce the necessity of a comprehensive diagnostic approach, leveraging both CT and MRI to ascertain the soft tissue characteristics and the exact anatomical delineation of the defect, thus ensuring an accurate diagnosis and averting unwarranted surgical interventions. Moreover, the ability to differentiate Stafne's cavity from other osseous lesions with a pseudocystic phenotype is of paramount importance, as several other odontogenic and non-odontogenic radiolucencies may present with radiographic similarities to this depression. In light of the benign nature of Stafne's cavity, and the non-requirement for surgical intervention, it's pivotal that clinicians maintain a high degree of vigilance and nosological precision to prevent misdiagnosis and avoid unnecessary surgical or dental procedures.

The authors declare that no conflict of interest is present for this paper.

We confirm that the manuscript has been thoroughly reviewed and approved by all the authors listed. We acknowledge that the requirements for authorship, as outlined in this document, have been satisfactorily fulfilled, and each author affirms that the manuscript reflects diligent and honest research work.

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