

# Anterior Lingual Mandibular Bone Cavity as a Diagnostic Challenge: Two Case Reports

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In 1942, Stafne was the first to report the presence of bone cavities in 35 patients.<sup>1</sup> These usually create a well-demarcated radiolucency in the posterior mandibular angle and are synonymously known under different names in the literature: Stafne bone cyst, static bone cavity, latent bone cyst, lingual mandibular bone cavity, mandibular embryonic defect, idiopathic bone concavity of the mandible, and developmental submandibular gland defect of the mandible. These cavities occur in the area between the mandibular first molar and the mandibular angle below the mandibular canal outline. Other than this radiographic presence, they have no real clinical presentation. They are innocuous convexities in the lingual cortex of the mandible that would be of no consequence if they did not appear as radiolucency. It may be difficult to distinguish them from more serious conditions such as cysts and benign or malignant tumors. Surgical exploration has shown that this hard

tissue configuration represents a focal concavity of the cortical bone on the lingual side of the mandible. In most cases, regular salivary gland tissue was the histologic finding.<sup>2</sup> This suggested a developmental origin: 1 theory was that part of the submandibular gland was entrapped in the lingual mandibular cortex. Muscle, fibrous connective tissue, blood vessels, fat, or lymphoid tissue were also found in some cases.<sup>3</sup> Stafne bone cavity is not considered extremely rare. The incidence ranges from 0.1% to 1.3%.<sup>4</sup>

The first report of Stafne bone cavity in the premolar region was issued by Richard and Ziskind in 1957.<sup>5</sup> The sublingual gland or aberrant salivary gland tissue has also been associated with this bony cavity in the anterior mandible.<sup>6</sup>

In the anterior mandible, the mandibular canal as an anatomic landmark for identifying latent bone cysts is absent. Thus, a diagnosis of Stafne bone cavity is difficult on plain radiographs; and endodontic treatment, bone trephining, and bone exploration may result from an incorrect diagnosis.<sup>7,8</sup> Cone-beam computed tomography, a fast developing 3-dimensional imaging technique, is becoming increasingly available in dental practice, being used most frequently in the planning of dental implants. Even with a low radiation dose, the quality of the hard tissue image is comparable to spiral computed tomograms in the bone window. However, soft tissues cannot be distinguished from each other.

Our objective is to describe 2 new cases of Stafne bone cavity in the anterior mandible and to discuss the differential diagnosis process. We introduce cone-beam computed tomography as a suitable noninvasive diagnostic and follow-up modality for this bony configuration in the anterior mandible.

## Case 1

A 58-year-old male patient consulted our outpatient clinic after his dentist noticed a sharply bordered unilateral radi-

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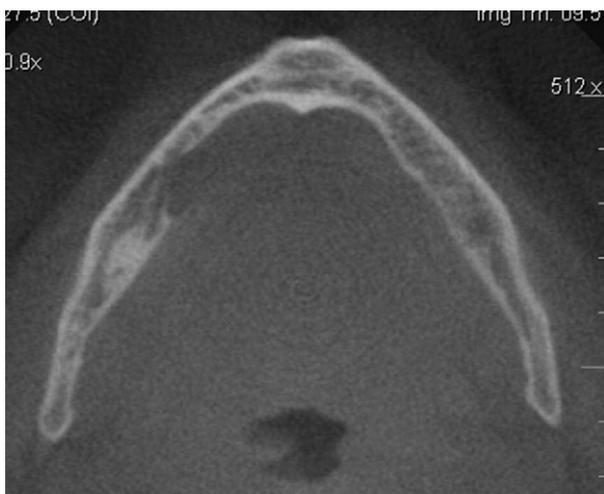
**FIGURE 1.** Panoramic radiograph demonstrating a sharply bordered unilateral radiolucency in the mandible.

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olucency in the mandible. The patient's history was not significant and the lesion was asymptomatic. The lesion was located in an edentulous zone between the lower right canine and the first molar and appeared to have no relation to the adjacent teeth (Fig 1). However, the 1.5- × 3-cm lesion in the projection of the mandibular canal seemed to be associated with the same. On cone-beam computed tomogram a lingual osteolysis was suspected, interrupting the lingual compact bone and affecting the mandibular canal with only poorly defined margins (Fig 2). Because a neoplasm could not be completely excluded, a biopsy was taken under general anesthesia. Histologic examinations showed a mixed salivary gland tissue with a slight chronic infection in the area of the ducts and periductal fibrosis.

## Case 2

A 50-year-old female patient was referred to the outpatient clinic of the department of oral and maxillofacial surgery with a suspect radiolucency in the region of the left lower canine. A few weeks previously, the radiolucency was found on a panoramic radiograph during routine dental



**FIGURE 2.** Lingual defect demonstrated in the axial view on cone beam computed tomogram.

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**FIGURE 3.** Panoramic radiograph illustrating another anterior radiolucency.

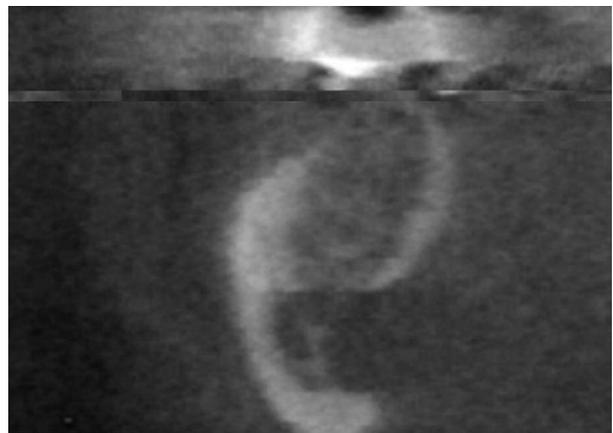
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examination (Fig 3). The patient's dental and medical histories were uneventful. No older radiographs were available for comparison. A periapical radiograph was obtained. The 20- × 15-mm lesion appeared to relate to the adjacent tooth, which was asymptomatic. There were no abnormal findings and all teeth in the region responded within normal limits to the temperature pulp test. The differential diagnosis was osteoporotic focal defect of the marrow, apical granuloma, residual cyst, or latent bone cyst. An occlusal radiograph and ultrasound did not provide further information, so cone-beam computed tomography was performed (Fig 4).

The radiologic examination showed a 20-mm-wide and 15-mm-long bony impression on the lingual side of the anterior mandible, which did not have any relation to an adjacent tooth. A follow-up radiograph taken 5 months later did not show any alteration of the radiolucency. Therefore, no invasive therapy was performed on this patient.

## Discussion

Ectopic salivary gland tissue has been described on many sites within the head and neck region including the lateral and posterior neck, tongue, middle ear



**FIGURE 4.** Coronal plane of the Stafne cavity.

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thyroid, pituitary gland, and mandible. The inclusion of salivary gland tissue in the mandible is a rare phenomenon, which is most commonly seen in the posterior region and more unusually in the anterior mandible. According to de Courten et al the incidence in the anterior part of the mandible is approximately 0.009% to 0.03%.<sup>2</sup> Male patients seem to be more often affected (approximately 80%), with a peak in their fifth and sixth decades.<sup>9-11</sup> Only 40 cases have been reported in the English-language literature and 15 in other languages to this date. Thirty-two of the reported cases are of archeological dried skull specimens.<sup>2,4,11,12</sup>

The pathogenesis of these lesions is not fully understood. Hypotheses put forward suggest that the defect is congenital or develops through pressure resorption. The first theory suggests that part of the salivary gland becomes entrapped during the development and ossification of the mandible.<sup>12,13</sup> Because Stafne lesions are much more common in adults, it also seems possible that they develop later in life due to ossification of the mandible. This hypothesis is supported by the fact that histologic studies have shown disruptions of the mandible's increment lines and the inferior border of the mandible in some cases.<sup>14</sup> Most lesions are asymptomatic. On rare occasions, the lingual defect can be clinically palpated. It has been established that such lesions are nonprogressive.<sup>12</sup>

Clinically and radiographically these entities can present diagnostic difficulties.<sup>11</sup> They are mostly asymptomatic. However, not all bone cavities will be well-demarcated impressions with a sclerotic border. They may be more irregular in their borders and correctly suggest a neoplasm. A sublingual impression may be represented as an ovoid or round radiolucency without a sclerotic border as seen in the present cases. Because a relation to the dental roots on plain radiographs is frequent in the anterior mandible, the differential diagnosis includes a periapical granuloma or radicular cyst; equally a keratocyst, an odontogenic tumor, or an idiopathic bone cavity may be unusual but possible considerations.<sup>9</sup>

A Stafne cavity found coincidentally in the posterior mandible is usually easy to diagnose because the localization and radiographic appearance are characteristic.<sup>15</sup> Any doubts can be eliminated with sialographic examinations of the submandibular gland.

Although sialography is helpful in doubtful posterior cases, it is impracticable and rarely effective in anterior cases due to the presence of many accessory Bartholin ducts adjoining the sublingual gland in the anterior mouth floor so that it gives little information on the submandibular gland.<sup>16</sup> Associated risks are duct trauma, acinar rupture, and a high failure rate.

Several investigators have described the use of computed tomography or magnetic resonance imaging

to specify the diagnosis, especially in the anterior case.<sup>3,12,17</sup> A computed tomogram offers a fast alternative because it can show the bony structure, soft tissue, and extension. The advantage of magnetic resonance imaging is better visualization of the soft tissue; however, its use is limited due to the high cost.

We recommend cone-beam computed tomography—a rapidly developing radiographic 3-dimensional imaging technique—to confirm the diagnosis. In contrast to conventional spiral computed tomography, the radiation dose ranges from 68 to 1,073  $\mu\text{Sv}$ , whereas computed tomography with a similar field of view produces 860  $\mu\text{Sv}$ .<sup>18</sup> The image quality of bony structures is comparable. Metal artifacts in particular are less intense. Admittedly, the volume of the examination is limited and soft tissues are not distinguishable from each other.

If a lingual impression in the anterior mandible produces a characteristic radiographic pattern with no clinical symptoms or other pathology, it may be diagnosed by an evaluation of radiographs. Inflammatory dental disease should be excluded. In most cases, no histopathology should be required and no treatment is indicated if 3-dimensional radiologic screening shows a Stafne cavity. Nevertheless, regular follow-ups are recommended to identify any changes.

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## Glomangioma: A Case Presentation

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The glomus body is an apparatus between the arterial and venous system located in the skin. It is encapsulated and works as a shunt between the arterioles and the venous blood. This is called the Sucquet-Hoyer canal.<sup>1</sup> Its purpose is to regulate the temperature in the skin. Occasionally, the glomus apparatus develops neoplasms. The glomus tumor is an entity that often appears as a slow growing, small, blue-red nodule that, clinically, might be reminiscent of a hemangioma. They are often approximately 5 mm in diameter, painful, and, in the fingertips, associated with a triad of symptoms: hypersensitivity to cold, paroxysmal pain, and pinpoint pain.<sup>1</sup> In the oral cavity, these symptoms seem to be more or less absent. Most glomus tumors are solitary but can also present as multifocal familial lesions. The glomus tumor is a rare neoplasm that is mostly found in the distal part of the extremities. To our knowledge, only 21 cases in the oral cavity have been reported.<sup>2-22</sup> No more than 20 cases of malignancy in the whole body have been reported,<sup>23</sup> with only 1 oral case representing a metastasis.<sup>10</sup>

Typical glomus tumors have varying proportions of glomus cells, vascular structures, and smooth muscle tissue, according to which they are grouped as solid

glomus tumors, glomangiomas, and glomangiomyomas.<sup>23,24</sup> The presented case, to date, occurred in one of the youngest patients and is one of only 4 glomangiomas convincingly presented in published studies.

### Report of a Case

An 11-year-old girl was referred to the Department of Oral and Maxillofacial Surgery at the Central Hospital, Eskilstuna, Sweden, in October 2000. The patient's parents had noted a well-defined, painless discoloration of the lower lip measuring 3 mm in diameter (Fig 1). The lesion was soft on palpation. The patient denied any other similar lesions throughout the body and was otherwise in good health. There was no record of heredity. Clinically, the lesion was reminiscent of a hemangioma. The lesion was removed with the patient under local anesthesia (Xylocaine with adrenaline 20 mg/mL, Astra, Södertälje, Sweden) with minimal margin, and the wound was sutured. The postoperative period was uneventful, and at 7 years postoperatively, no signs of recurrence had been seen (Fig 2).

### Histopathologic Findings

The specimen was fixed in 10% formalin and sectioned in 3- $\mu$ m sections that were stained with hematoxylin and eosin. The sections were also immunostained with antibodies to epithelial membrane antigen, pan-cytokeratin, vimentin, smooth muscle actin, and S-100.

The sections showed oral mucosa with several dilated vascular structures in the lamina propria and extending into the submucosa. The venous vessels were lined by a nondescript endothelium surrounded by layers of glomus cells (Figs 3, 4). The glomus cells were roundish with lightly stained cytoplasm and cytologically uniform, oval nuclei (Fig 5). Mitoses were virtually absent. Immunostaining showed a distinct expression of smooth muscle actin in the tumor

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