Developmental Mandibular Salivary Gland Defect

The Importance of Clinical Evaluation

Authored by Sako Ohanesian, DDS

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Developmental Mandibular Salivary Gland Defect: The Importance of Clinical Evaluation

LEARNING OBJECTIVES:

After reading this article, the individual will learn:

• to differentiate a developmental salivary gland defect from traumatic bone cysts and other lesions.
• to recognize the clinical and radiographic appearance of a developmental mandibular salivary gland defect (the intention is to avoid unnecessary biopsy).

ABOUT THE AUTHOR

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INTRODUCTION

A developmental mandibular salivary gland defect (also known as static bone cyst, static bone defect, Stafne bone cavity, latent bone cyst, latent bone defect, idiopathic bone cavity, developmental submandibular gland defect of the mandible, aberrant salivary gland defect in the mandible, and lingual mandibular bone concavity) is a deep, well-defined depression in the lingual surface of the posterior body of the mandible. More precisely, the most common location is within the submandibular gland fossa and often close to the inferior border of the mandible. In developmental bone defects investigated surgically, an aberrant lobe of the submandibular gland extends into the bony depression.

First recognized by Dr. Edward Stafne in 1942, numerous cases of developmental mandibular salivary gland defect have since been reported, and the lesion should not be considered rare.¹ In a study of 4963 panoramic images of adult patients, 18 cases of salivary gland depression were found by Karmiol and Walsh², an incidence of nearly 0.4%. Most authorities now agree that this entity is a congenital defect, although it has rarely been observed in children and its precise anatomic nature is still uncertain. Also unexplained is the fact that far more cases have been reported in men than in women.³

The lesion, usually asymptomatic and discovered during routine radiographic examination, appears as an ovoid radiolucency, generally situated between the mandibular canal and the inferior border of the mandible, just anterior to the angle. Rare examples are located in the apical region of the mandibular premolars or cuspids, and are related to the sublingual gland fossa. The margins of the radiolucent defect are well-defined by a dense radiopaque line. This cortical margin is usually thicker on the superior aspect. This appearance is the result of the x-rays passing tangentially through the relatively thick walls of the depression. It is occasionally bilateral. The radiolucent defect may represent either actual enclavement of salivary gland tissue within the mandible during embryonic development or, more frequently, an indentation on the mandible with a portion of the submaxillary gland lying within the defect. Salivary gland defects are presumed to form by the remodeling of the mandibular cortex around an extension of salivary tissue. This theory is supported by findings of radiolucencies in association with each of the 3 salivary glands. Most surgical series have noted salivary tissue within the bony defect, but muscle, lymphatic tissue, and blood vessel have also been reported.
The lesion may be regarded properly as a developmental defect rather than a pathologic lesion. Histologically, normal salivary tissue is found, and no treatment is required except routine radiographic follow-up. It can and should be differentiated from the traumatic bone cyst (also referred to as hemorrhagic bone cyst). The traumatic bone cyst is an uncommon, unlined cavity of the jaws. Clinically, the lesion is asymptomatic in the majority of cases and is often accidentally discovered on routine radiological examination. Pain is the presenting symptom in 10% to 30% of the patients. Other, more unusual symptoms include tooth sensitivity, paresthesia, fistulas, delayed eruption of permanent teeth, displacement of the inferior dental canal, and pathologic fracture of the mandible.\(^4\)\(^-\)\(^6\)

Expansion of the cortical plate of the jawbone is often noted, usually buccally, resulting in intraoral and extraoral swelling and seldom causing deformity of the face. On radiological examination, a traumatic bone cyst usually appears as a unilocular radiolucent area with an irregular but well-defined (or partly well-defined) outline, with or without sclerotic lining around the periphery of the lesion. The traumatic bone cyst almost invariably lies above the mandibular canal on the intraoral periapical roentgenogram, while the salivary gland depression lies below the canal. Nevertheless, definitive differential diagnosis from other lesions sometimes cannot be made without surgical exploration.

It has been recognized that a sublingual salivary gland depression or inclusion may occur on the lingual surface of the anterior segment of the mandible. These asymptomatic lesions have generally appeared on the roentgenogram as a rather poorly circumscribed radiolucency in a location between the central incisor and first premolar area. They are far less common than the posterior lesion. A complication occasionally reported in the literature is the development of a true central salivary gland neoplasm from the included salivary gland tissue, but this is rare.\(^7\)

### CASE REPORT

The patient in this case was a white 36-year-old male, with failed endodontic therapy involving tooth No. 19. The patient was healthy (ASA I), did not report any relevant information regarding medical or dental history, and did not mention the use of any medication. Expansion of the mandible and hydration of mucous membrane were normal. Endodontic consultation confirmed root fracture, and the tooth was extracted and replaced with a root form implant. The option of a 3-unit bridge was given to patient. The radiograph disclosed a well circumscribed radiolucency inferior to the mandibular canal and located in the region of the right mandibular second and third molars (Figures 1 and 2). The diameter measured approximately 2 cm. No symptoms were reported.

[Figure 1. CT slices/panoramic views showing a well-defined radiolucent lesion in the right mandible.]

[Figure 2. Lesion in standard panoramic x-ray and implant placement.]
On 3-dimensional imaging views of the lingual aspect of the mandible obtained with a NewTom 3D cone beam CT scanner (AFP Imaging Corp), it was observed that this radiolucency represented a cortical indentation or depression (Figure 3). A diagnosis of Stafne bone cavity was made, and no further therapy was instituted. The pathologist's recommendation was to simply observe the area radiographically in the event that it became enlarged and would necessitate a surgical biopsy.

**DISCUSSION**

Many terms have been used to describe asymptomatic radiolucencies at the angle of the mandible. Similar defects related to the sublingual and parotid glands have been described, located at the mandibular symphysis and the mandibular rami, respectively. Some researchers apply the term Stafne bone cyst to lesions associated with any of the salivary glands, while others restrict the term to the submandibular gland, preferring more specific terms such as anterior lingual mandibular salivary gland defect for the sublingual gland.

Dental professionals are facing an ever-increasing emphasis on a thorough clinical examination of each patient. As a result, the dentist is often confronted with the need to further evaluate any deviation from normal, including the decision to biopsy a suspected lesion. To avoid any unnecessary procedures and treatments, it is important to be aware of the existence of other anatomic variations in the examination process. Awareness of these entities can save the patient from unnecessary invasive procedures.

Most case reports of Stafne bone cavities have discussed the findings on intraoral dental films, plain films of the mandible, or orthopantographs. Although these imaging techniques are often sufficient for diagnosis, they may not be definitive when the lesion is atypical. In these situations, confirmatory testing is warranted, as the differential diagnosis for mandibular radiolucencies includes traumatic bone cyst, periapical cyst, dentigerous cyst, odontogenic keratocyst, nonossifying fibroma, fibrous dysplasia, ameloblastoma, giant cell tumor, focal osteoporotic bone marrow defect, basal cell nevus syndrome, and brown tumor of hyperparathyroidism. (See Table. pages 4 and 5)

**CONCLUSION**

Given the possible clinical presentation of the various lesions described, it is important for the dentist to be aware of the existence of these anatomic variations in the examination process. Cystic-appearing lesions that occur in the mandible are often difficult to distinguish from one another with radiography. They are all usually benign, but some can be locally aggressive and destructive. The patient history and careful consideration of the location of the lesion within the mandible, its borders, its internal architecture, and its effects on adjacent structures generally make it possible to narrow the differential diagnosis. Awareness of these entities can save the patient from unnecessary treatment and unwarranted procedures.
## Clinical Presentation

Peaks in second decade; usually in body of mandible; painless in most cases; swelling noted in one fourth of cases.

Diagnosis

- Classically defined radiolucency; margins may be uneven but clear; may extend between tooth roots creating a scalloped pattern.

Radiographic appearance:
- Clinical finding of an empty bony space (pseudocyst); collagen and fibrin line the dead space; bony spine may be noted along the bony margin.

Radiographic Findings

- Radiographic appearance; clinical finding of an empty bony space (pseudocyst); collagen and fibrin line the dead space; bony spine may be noted along the bony margin.

Treatment

- Surgical exploration; observation for resolution.

- Excellent; small risk of recurrence.

Table. Comparison of Various Lesions That Can Be Confused With Static Bone Defect.

<table>
<thead>
<tr>
<th>Lesion</th>
<th>Etiology</th>
<th>Clinical Presentation</th>
<th>Radiographic Findings</th>
<th>Diagnosis</th>
<th>Treatment</th>
<th>Prognosis</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Traumatic bone cyst</strong></td>
<td>Unknown in most cases; may be due to traumatic injury producing inflammatory hemorrhage and subsequent cell necrosis; alternative theory suggests degeneration of primary intrabony pathology.</td>
<td>No symptoms; discovered incidentally.</td>
<td>Round to oval; radiolucent area, above or below the tooth root.</td>
<td>Radiographic appearance.</td>
<td>Recognition only.</td>
<td>Excellent.</td>
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<td><strong>Mandibular salivary gland defect (Static bone cavity)</strong></td>
<td>Developmental depression of the lingual side of the mandible; the absent ramus of the submandibular salivary gland and/or adipose tissue fills the body of mandible; depression creates problems similar to radiographic findings.</td>
<td>No symptoms; discovered incidentally.</td>
<td>Round to oval; radiolucent area, above or below the tooth root.</td>
<td>Radiographic appearance.</td>
<td>Recognition only.</td>
<td>Excellent.</td>
</tr>
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<td><strong>Periapical cyst</strong></td>
<td>A radicular cyst that most likely results when roots of epithelial cells in the periodontal ligament are stimulated by inflammatory products from a nonvital tooth.</td>
<td>Often, periapical cysts do not produce symptoms unless secondary infection occurs.</td>
<td>Located approximately in the apex of a nonvital tooth; occasionally, appear on the mesial or distal surface of a tooth root, at the opening of an accessory canal, or intraorally in a deep periodontal pocket; most (85%) found in the maxilla.</td>
<td>Radiographic appearance.</td>
<td>Treatment of a tooth with a radicular cyst may include extraction, endodontic therapy, and surgical surgery, treatment of a large cyst usually involves surgical removal or marsupialization.</td>
<td>Excellent; recurrence is unlikely if removed completely.</td>
</tr>
<tr>
<td><strong>Dentigerous cyst</strong></td>
<td>A developmental odontogenic cyst arising subsequent to separation between dental follicle and the crown of an associated unerupted tooth; proliferation of reduced enamel epithelium lining the follicle, with fluid accumulation between epithelium and impacted tooth crown; degeneration of the stellate reticulum component of enamel organ occurs during odontogenesis.</td>
<td>Most commonly involves frequently impacted teeth: mandibular third molars, followed by maxillary canines; usually rooted during second and third decades; asymptomatic; discovered incidentally; on routine radiographic examination; painless; jawular expansion may occur; cysts are thin and rarely perforated.</td>
<td>Well-defined radiolucency; enclosed crown of unerupted tooth; corticated/palqueom margins unless infected; may produce root resorption of adjacent erupted teeth; usually unicellular; less commonly multicellular.</td>
<td>Radiographic features.</td>
<td>Excise with curvilinear bony continuity.</td>
<td>Excellent; recurrence rate varies from 10% to 30% (greatest in patients with a syndrome).</td>
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<td><strong>Odontogenic keratocyst</strong></td>
<td>A benign, aggressive developmental odontogenic cyst; may be associated with mutation of PTCH tumor suppressor gene.</td>
<td>5% to 10% of nascent keratocysts; usually occur sporadically as an isolated finding; about 5% are associated with nevoid basal cell carcinoma, 5% of patients have multiple odontogenic keratocysts (OMCs) and no syndrome.</td>
<td>Can occur in any area of maxilla or mandible; rarely may arise in gingival soft tissue only; mandible is preferred site in 65% to 79% of cases; often seen in a sagittal relationship; discrete keratocyst; usually in relation to teeth; may be unicellular or multicellular.</td>
<td>Radiographic features.</td>
<td>Excise with curvilinear bony continuity.</td>
<td>Excellent; recurrence rate varies from 10% to 30% (greatest in patients with a syndrome).</td>
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<td><strong>Nonossifying fibroma</strong></td>
<td>Unknown in most cases; lesions occur as a result of developmental alterations at the epithelial plate; not neoplasms, but developmental defects, tend to occur after the age of 2, a muscle pull and peridental injury may be a contributing factor.</td>
<td>Majority of all NOFs are asymptomatic and are discovered incidentally on radiographs; symptomatic lesions may progress with mild pain and swelling of short duration; may have bone tenderness with palpation.</td>
<td>On plain film radiographs, NOFs appear as eccentric, radiolucent, well-corticated masses; lesions may extend into the mandibular cavity; long axis of the NOF is most commonly seen parallel to the long axis of the bone and are usually located medially.</td>
<td>Radiographic features.</td>
<td>Excellent; unlikely if removed completely.</td>
<td>Generally excellent.</td>
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<tr>
<td>Fibrous dysplasia</td>
<td>Clinical Presentation</td>
<td>Radiographic Findings</td>
<td>Diagnosis</td>
<td>Treatment</td>
<td>Prognosis</td>
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<td>Unknown in most cases; skeletal abnormalities constitute the cardinal feature; the condition is often monoosseous, but may be polyosseous. Fibrous dysplasia is of greater concern to the dentist due to frequency with which jaw bones are affected; nearly every bone has been reported involved.</td>
<td>Occurs with equal predilection for males and females; more common in children and young adults, painless swelling or bulging of the jaw; swelling usually involves the labial or buccal plate, seldom the lingual aspect; possible malalignment, tipping or displacement of teeth, excess is almost invariably intact after the lesion.</td>
<td>Generally a small unicocular radiolucency or a somewhat larger multicellular radiolucent cyst; both with a rather well-circumscribed border and containing a network of fine bony trabeculae. Increased trabeculation could render the lesion more opaque; the periphery of lesions most commonly is ill defined, with a gradual blending of normal trabecular bone into an abnormal trabecular pattern.</td>
<td>Monostatic fibrous dysplasia is often discovered as an incidental radiographic finding. Patients with jaw involvement first may complain of abnormal facial swelling or an altering deformity of the alveolar process; pain and pathologic fractures are rare. If extensive cortical lesions have occurred on serve timeframe, neurologic symptoms such as anosmia, deafness, or blindness may develop.</td>
<td>Surgical removal of lesion.</td>
<td>Majority of lesions are too large at the time of original diagnosis to evolve successfully without leaving facial deformity or, in the case of the mandible, weakening of the bone so as to invite pathologic fracture. Numerous cases reported in which monostotic fibrous dysplasia has undergone spontaneous malignant transformation into sarcoma.</td>
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Anameloblastoma

A benign, aggressive jaw tumor of odontogenic epithelial-endothelial origin; the most common odontogenic tumor after the odontoma; incidence of 0.3 cases per million people.

Peak incidence during third to fifth decades; 80% occur in the mandible, chiefly in maxilla and anterolateral region, often present with association with unerupted third molar teeth; may produce marked deformity, facial asymmetry; periphery variant occurs in gingival tissue of older adults 95% to seventh decades; slow growing, but persistent.

Osseous or radioulucent with smooth, even borders; may be unencapsulated or multilocular; root resorption or tooth displacement may be seen, can expand affected jaw, cortical perforation may occur.

Shunts, stones, islands of odontogenic epithelium; peripheral layer of cuboidal to columnar anameloblastic-like cells enclosing a cell population analogous to odontoblastic reticulum of the enamel organ; several histologic patterns described have no clinical relevance; malignant variants rarely seen.

Varies with subtype, size, and location; solid/multilocular lesions generally require local excision or resection, cystic variant requires local excision via curettage may follow curettage only.

Generally good, recurrence rates higher with conservative treatment, recurrence rate up to 15% following marginal resection; long-term follow-up necessary.

Giant cell tumor

Probable reactive or reparative process; speculation suggests it may represent a developmental anomaly.

Bone expansion; most cases in those less than 30 years of age; female predominance; rare exclusively in mandible or maxilla—very rarely in facial bones, occasionally in mandible; prominent 2.5 to 7.1 cm over that in maxilla; usually anterior to molar teeth; most cases are slow growing, minimally symptomatic, and asymptomatic; some cases are recurrent and exhibit aggressive behavior with pain, perforation, and rapid enlargement; no radiographic or histologic features can be used to separate nonaggressive lesions from aggressive lesions.

Usually multilocular, occasionally unicellular, radiolucency; margins are usually well defined, borders may be ill defined; often involves areas of low density; fluctuate in size.

Intracranial/basal primary hyperparathyroidism should be ruled out.

Intratumoral vessels; primary hyperparathyroidism should be ruled out.

Through curettage; marginal reaction; aggressive or recurrent. Caution:勿on surgical approach, recurrence rate up to 25% following marginal resection; long-term follow-up necessary.

Aggressive variant has high recurrence rate; generally good.

Focal odontogenic bone marrow defect

Etiology is unknown but has been postulated to be bone marrow hyperplasia, persistent embryonic marrow remnants, or site of abnormal healing following extraction, trauma, or local irritation.

In reported cases, 77% occurred in women, and they involved the mandible in 85% of cases, asymptomatic and discovered only during routine roentgenographic examination.

Leukemia has a predilection for the mandibular marrow area; generally appears as a radiolucency of variable size, a few millimeters to a centimeter or more, with a poorly defined margin, indicative of lack of reactivity of adjacent bone; most common in mandibular areas, suggesting they result from failure of normal bone regeneration after tooth extraction.

The reantgenographic appearance of the focal odontogenic bone marrow defect of the jaw is not unlike that of residual dental infections, central neoplasms, or even the traumatic cyst of bone.

Recognition only; when doubt exists about the true nature of the radiolucency, a histologic study with findings at 3-month intervals may be prescribed; the maxillary space should not increase in size.

Good.

Brown tumor syndrome

A hereditary condition, transmitted as an autosomal dominant trait, with high penetrance and variable expressivity.

Complex syndrome which includes a variety of possible abnormalities including dental and maxillofacial anomalies such as odontogenic keratocysts and early mandibular prognathism.

Multiple keratocysts may develop bilaterally and can vary in size from one mm to several centimeters in diameter; a radiopaque line of the calcified keratocyst border may be present on the posteroanterior skull projection; occasionally the calcification may appear leptomeningeal.

Starts to appear early in life, usually after 5 years of age and before 30, with development of jaw cysts and bone involvement; jaw cysts are common, facial abnormalities occur in multiple quadrants; the presence of oral hypoplasia and other cystic characteristics differentiates basal cell nevus syndrome from other abnormalities characterized by multiple keratocystic lesions.

High recurrence rate of the keratocysts associated with this syndrome; several cases of ameloblastomas have developed in cysts, thus emphasizing the importance of surgical removal of the cysts and its histologic examination.

It is reasonable to examine the patient yearly for new and recurrent cysts; a panoramic film serves as an adequate screening film; referral for genetic counseling may be appropriate.

Brown tumor in hyperparathyroidism

May appear in any bone, but are frequently found in the facial bones and jaws; these lesions may be multiple within a single bone.

Varially defined margins and may produce cortical expansion; if solitary, tumor may resemble a central giant cell granuloma, therefore, if giant cell granuloma occurs later than the second decade, the patient should be screened for an increase in serum calcium, PTH, and alkaline phosphatase.

Occasionally peripheral radiographs reveal loss of the lamina dura in patients with hyperparathyroidism; loss of lamina dura may occur around one tooth or all the remaining teeth.

Manifestations cover a broad range, but most patients have renal calculi, psychotic problems, or bone and joint pain; gradual loosening, drifting, and loss of teeth may occur; because of calcium overload, the serum calcium level should be checked at different intervals; the serum alkaline phosphatase level may be elevated in hyperparathyroidism.

Surgical removal; the site of brown tumor heals with bone that is radiographically more sclerotic than normal.

After successful surgical removal of the causative parathyroid adenoma, almost all radiographic changes revert to normal; the site of a brown tumor often heals with bone that is radiographically more sclerotic than normal.
REFERENCES

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POST EXAMINATION QUESTIONS

3. Clinical presentation of the salivary gland defect is:
   a. asymptomatic—discovered incidentally.
   b. painless swelling.
   c. bulging of the jaw.
   d. painful swelling.

4. Incidence rate of Stafne bone defect is nearly:
   a. 0.4%.
   b. 4%.
   c. 40%.
   d. 0.1%.

5. The developmental salivary gland defect is best described as:
   a. benign neoplasm.
   b. variations of normal.
   c. cyst.
   d. a premalignant lesion.

6. Differential diagnosis for mandibular radiolucencies may include:
   a. dentigerous cyst.
   b. traumatic bone cyst.
   c. odontogenic keratocyst.
   d. all of the above.

7. Size of the Stafne bone defect usually ranges between:
   a. 3 to 5 mm.
   b. 1 to 3 mm.
   c. 3 to 5 cm.
   d. 1 to 3 cm.

8. Which of the following techniques are useful in diagnosing the Stafne bone cyst?
   a. periapical x-ray
   b. panoramic x-ray
   c. cone beam CT imaging
   d. all of the above
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