



Developmental Mandibular Salivary Gland Defect

The Importance of Clinical Evaluation



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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 pan-

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oramic images of adult patients, 18 cases of salivary gland depression were found by Karmioli 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.

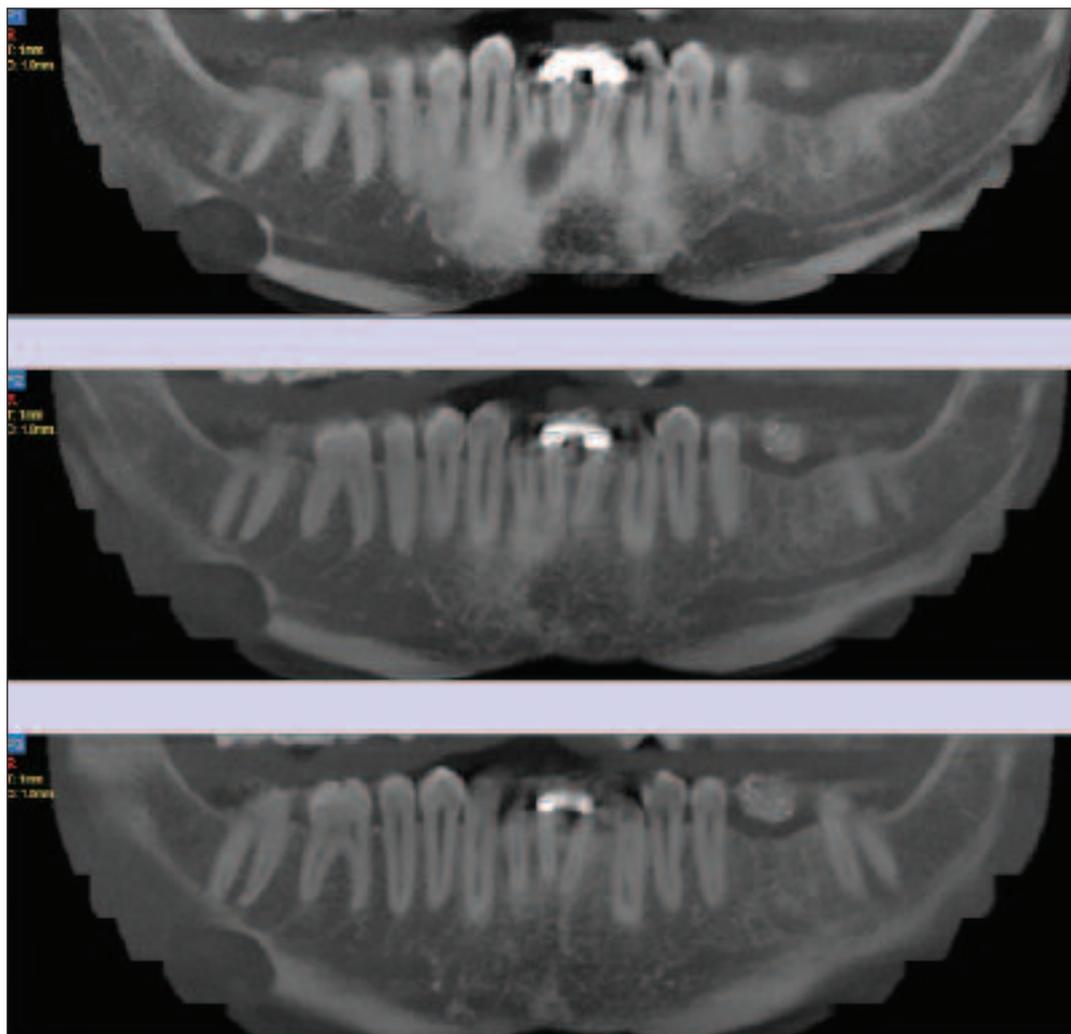


Figure 1. CT slices/panoramic views showing a well-defined radiolucent lesion in the right mandible.

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 hemor-

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Table. Comparison of Various Lesions That Can Be Confused With Static Bone Defect. 7,12,13

	Etiology	Clinical Presentation	Radiographic Findings	Diagnosis	Treatment	Prognosis
Traumatic bone cyst	Unknown in most cases; may be due to traumatic injury producing intramedullary hemorrhage and subsequent clot resorption; alternative theory suggests degeneration of primary intrabony pathology.	Peaks in second decade; usually in body of mandible; painless in most cases; swelling noted in one fourth of cases.	Clearly 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; lamellar bone may be noted along the bony margin.	Surgical exploration; observation for resolution.	Excellent; small risk of recurrence.
Mandibular salivary gland defect (Stafne bone cavity)	Developmental depression of the lingual side of the mandible; the aberrant lobe of the submandibular salivary gland and/or adipose tissue fills the body of mandible defect; depression created produces characteristic radiographic findings.	No symptoms; discovered incidentally.	Round to ovoid radiolucency below inferior alveolar canal, above inferior border and below third molar area; well-defined by a dense hypercorticated margin; size range of one to 3 cm; rarely noted in premolar and canine areas.	Radiographic appearance.	Recognition only.	Excellent.
Periapical cyst	A radicular cyst that most likely results when rests of epithelial cells in the periodontal ligament are stimulated by inflammatory products from a nonvital tooth.	Often, periapical cysts do not produce symptoms unless secondary infection occurs.	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 infrequently in a deep periodontal pocket; most (60%) found in the maxilla.	A cyst that becomes large may cause swelling; the swelling may feel bony and hard if the cortex is intact, crepitant as the bone thins, and rubbery if bone destruction has occurred; outline of cyst is usually curved or circular unless influenced by surrounding structures such as cortical boundaries.	Treatment of a tooth with a radicular cyst may include extraction, endodontic therapy, and apical surgery; treatment of a large cyst usually involves surgical removal or marsupialization.	Excellent; recurrence is unlikely if removed completely.
Dentigerous cyst	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.	Most commonly involves frequently impacted teeth: mandibular third molars, followed by maxillary canines; usually noted during second and third decades; asymptomatic and discovered on routine radiographic examination; painless jaw/alveolar expansion may occur; cortex is thinned and rarely perforated.	Well-defined radiolucency enclosing crown of unerupted tooth; corticated/opaque margins unless infected; may produce root resorption of adjacent erupted teeth; usually unilocular; less commonly multilocular.	Cysts without secondary inflammation are thin, cuboidal, nonkeratinized epithelial lining 2 cell layers thick with flat epithelial-connective tissue interface; loosely arranged collagen bundles; cysts with secondary inflammation have hyperplastic, nonkeratinized squamous epithelial lining with epithelial ridge development; variable chronic inflammatory cell infiltrate within condensed collagen stroma.	Cyst enucleation and extraction of associated tooth; marsupialization prior to excision may be considered if very large.	Excellent; possible complications include: pathologic fracture with large lesions and neoplastic transformation of epithelial lining.
Odontogenic keratocyst	A benign, aggressive developmental odontogenic cyst; may be associated with mutation of PTCH tumor suppressor gene.	5% to 15% of odontogenic cysts; usually occurs sporadically as an isolated finding; about 5% are associated with nevoid basal cell carcinoma; 5% of patients have multiple odontogenic keratocysts (OKCs) and no syndrome.	Can occur in any area of maxilla or mandible; rarely may arise in gingival soft tissue only, mandible is preferred site in 65% to 78% of cases; often seen in a dentigerous relationship; discrete radiolucency, usually in relation to teeth; may be unilocular to multilocular.	Radiographic features.	Excision with curettage of bony confines.	Recurrence rate varies from 10% to 30% (greatest in patients with a syndrome).
Nonossifying fibroma	Unknown in most cases; lesions occur as a result of developmental aberrations at the epiphyseal plate; not neoplasms, but developmental defects; tend to occur after the age of 2, a muscle pull and periosteal injury may be a contributing factor.	Majority of all NOFs are asymptomatic and are discovered incidentally on radiographs; symptomatic lesions may present with mild pain and swelling of short duration; may have bone tenderness with palpation.	On plain film radiographs, NOFs appear as eccentric, multi or uniloculated, ovoid lesions in the metaphysis of bone with sclerotic margins; lesions may extend into the medullary cavity; long axis of the NOF is most commonly seen parallel to the long axis of the bone and are usually located medially.	Histologically, the lesions contain whorled bundles of connective tissue cells admixed with foamy histocytes, hemosiderin, hemorrhage, collagen, multinucleated giant cells, and bone trabeculae.	Treatment varies depending on the size and severity of the NOF; surgery is often not required to treat NOF due to a high rate of spontaneous regression and a lack of symptoms; symptomatic lesions should first be treated conservatively—conservative care consists of limited activity and immobilization, in addition to yearly or bi-yearly radiographs; curettage or bone grafting.	Generally excellent.

	Etiology	Clinical Presentation	Radiographic Findings	Diagnosis	Treatment	Prognosis
Fibrous dysplasia	Unknown in most cases; skeletal aberrations constitute the cardinal feature; the condition is often monostotic, but may be polyostotic; monostatic fibrous dysplasia is of greater concern to the dentist due to frequency with which jaws are affected; nearly every bone has been reported involved.	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; mucosa is almost invariably intact over the lesion.	Generally a small unilocular radiolucency or a somewhat larger multilocular radiolucency; 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.	Monostatic fibrous dysplasia is often discovered as an incidental radiographic finding; patients with jaw involvement first may complain of unilateral facial swelling or an enlarging deformity of the alveolar process; pain and pathologic fractures are rare; if extensive craniofacial lesions have impinged on nerve foramina, neurologic symptoms such as anosmia, deafness, or blindness may develop.	Surgical removal of lesion.	Majority of lesions are too large at the time of original diagnosis to excise surgically 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.
Ameloblastoma	A benign, aggressive jaw tumor of odontogenic epithelial ectodermal 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 molar and ramus region; often presents in association with unerupted third molar teeth; may produce marked deformity, facial asymmetry; peripheral variant arises in gingival tissue of older adults fifth to seventh decades; slow growing, but persistent.	Osteolytic or radiolucent with sclerotic, smooth, even borders; may be unilocular to multilocular; root resorption or tooth displacement may be seen; can expand affected jaw; cortical perforation may occur.	Sheets, strands, islands of odontogenic epithelium; peripheral layer of cuboidal to columnar ameloblast-like cells enclosing a cell population analogous to stellate reticulum of the enamel organ; several histologic patterns described have no clinical relevance; malignant variants rarely seen.	Varies with subtype, size, and location; solid/multicystic lesions generally require local incision or resection; cystic variant requires local excision as recurrences may follow curettage only.	Generally good, recurrence rates higher with conservative treatment; recurrence rates of up to 15% following marginal resection; long-term follow-up necessary.
Giant cell tumor	Probably reactive or responsive in nature; speculation suggests it may present a developmental anomaly.	Bony expansion; most cases in those less than 30 years of age; female predominance; near exclusivity in mandible or maxilla—rarely in facial bones; occurrence in mandible predominates 3:1 over that in maxilla; usually anterior to molar teeth; most cases are nonaggressive, slow growing, 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 unilocular, radiolucency; margins are usually well defined; borders may be scalloped; can displace teeth; wide-size variation at time of presentation.	Incisional biopsy; primary hyperparathyroidism should be ruled out.	Thorough curettage; marginal resection if aggressive or recurrent; Calcitonin may be successful in some cases; intralesional corticosteroid placement in small lesions may be successful.	Aggressive variant has high recurrence rate; generally good.
Focal osteoporotic bone marrow defect	Etiology is unknown but has been postulated to be bone marrow hyperplasia, persistent embryologic marrow remnants, or site of abnormal healing following extraction, trauma, or local inflammation.	In reported cases, 77% occurred in women, and they involved the mandible in 83% of cases; asymptomatic and discovered only during routine roentgenographic examination.	Lesion has a predilection for the mandibular molar area, generally appears as a radiolucency of variable size, a few millimeters to a centimeter or more, with a poorly defined periphery indicative of lack of reactivity of adjacent bone; most common in edentulous areas, suggesting they result from failure of normal bone regeneration after tooth extraction.	The roentgenographic appearance of the focal osteoporotic bone marrow defect of the jaws 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 longitudinal study with films at 3-month intervals may be prescribed; the marrow space should not increase in size.	Good.
Basal cell nevus 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 osseous anomalies such as odontogenic keratocysts and mild 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 falx cerebri may be prominent on the posteroanterior skull projection; occasionally the calcification may appear laminated.	Starts to appear early in life, usually after 5 years of age and before 30, with development of jaw cysts and skin, basal cell carcinomas; lesions occur in multiple quadrants; the presence of cortical boundary and other cystic characteristics differentiate basal cell nevus syndrome from other abnormalities characterized by multiple radiolucencies.	High recurrence rate of the keratocysts associated with this syndrome; several cases of ameloblastoma have developed in cysts, thus emphasizing the importance of surgical removal of the cysts and their 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.	Variably defined margins and may produce cortical expansion; if solitary, tumor may resemble a central giant cell granuloma, therefore, if a 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 peripical 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, peptic ulcers, psychiatric problems, or bone and joint pain; gradual loosening, drifting, and loss of teeth may occur; because of daily fluctuations, the serum calcium level should be tested 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.

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rhagic 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.

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

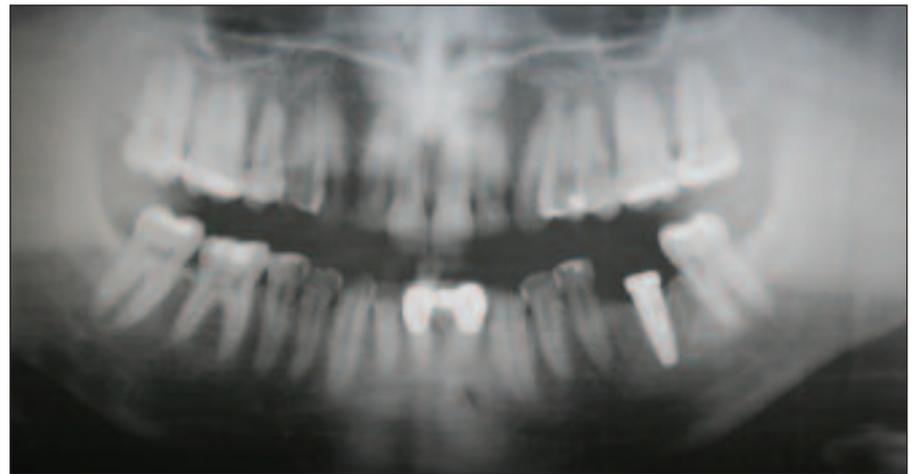


Figure 2. Lesion in standard panoramic x-ray and implant placement.

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.⁸⁻¹⁰ 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.¹¹

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 orthopantomographs. 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.^{7,12,13})

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

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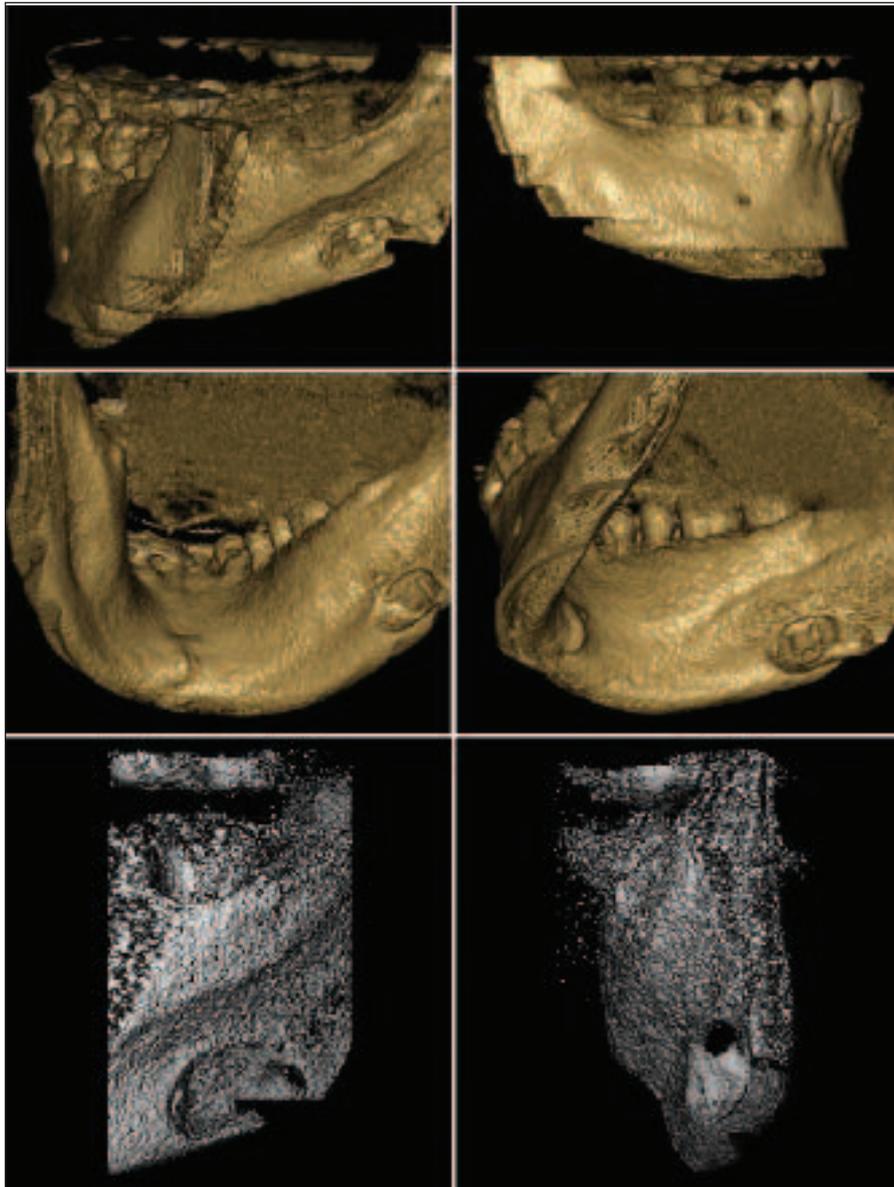


Figure 3. The 3-D images show a cortical depression.

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. ♦

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Disclosure: Dr. Ohanesian is not affiliated with NewTom Dental and has no financial interest in the company.

Continuing Education Test No. 98.2



To submit Continuing Education answers, use the answer sheet on page 122. On the answer sheet, identify the article (this one is Test 98.2), place an X in the box corresponding to the answer you believe is correct, detach the answer sheet from the magazine, and mail to Dentistry Today Department of Continuing Education.

The following 8 questions were derived from the article *Developmental Mandibular Salivary Gland Defect: The Importance of Clinical Evaluation* by Sako Ohanesian, DDS, on pages 116 through 121.

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).

1. A developmental salivary gland defect is located:
 - a. above the mandibular canal.
 - b. below the mandibular canal.
 - c. at the crest of the bone.
 - d. at the tuberosity.
2. Treatment for a salivary gland defect includes:
 - a. chemotherapy.
 - b. surgical intervention.
 - c. radiation.
 - d. none—recognition only.
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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