Asymptomatic intrabony radiolucency of the anterior mandible

Ho-Hyun Sun, DMD, MS, (Brian)[a,b] Sheng-Chuan Lin, DMD, (Charlie)[a,b]
Chan M. Park, DDS, MD, FACS,[a,b] and Jeffrey A. Elo, DDS, MS, FACS[c,d]
(Oral Surg Oral Med Oral Pathol Oral Radiol 2020;130:350−356)

CLINICAL PRESENTATION

A 33-year-old Hispanic female was referred to a large-scale, urban oral and maxillofacial surgery facility for evaluation of a mandibular radiolucency. The patient's medical history included allergy to penicillin and a diagnosis of mild depression. Her current medications included clindamycin and for third molar pericoronitis and sertraline for depression. She denied recent nausea, vomiting, fevers, or chills but reported occasional alcohol and marijuana use. Her surgical history was noncontributory, and she denied any current symptoms. There were no reports of night sweats, fatigue, or sudden weight loss.

Extraoral examination revealed a symmetric face and mildly limited mouth opening (approximately 35 mm) likely resulting from pericoronitis. The facial skin appeared intact, without apparent rashes or neurologic deficits. Palpation of the jaws and cervical regions did not reveal any signs of swelling, fluctuance, or tenderness. The cervical lymph nodes were unremarkable. Intraorally, she exhibited evidence of poor oral hygiene but no gross purulence or dental mobility. Bilateral mandibular vestibules and the floor of the mouth did not have any ulcerations, discrete white or red lesions, or swellings that could indicate an underlying pathology. All anterior teeth were vital to percussion, electric pulp testing, and cold.

Panoramic radiography was first performed as part of the screening protocol. The radiograph was compared with another panoramic radiograph that had been obtained approximately 13 months earlier and did not demonstrate any obvious pathology (Figure 1). The newly acquired image demonstrated an intrabony ovoid, well-defined, unilocular radiolucent lesion, measuring approximately 30×20 mm in size and positioned apically to her right mandibular incisors and canine (teeth #25, #26, and #27) (Figure 2). Three-dimensional cone beam computed tomography (CBCT) demonstrated intact lamina dura and bony cortications without apparent erosion or expansion (Figures 3 and 4). The well-defined outline of the lesion remained approximately 5 mm from the inferior border of the mandible, and its posterior border did not appear to encroach upon the mental foramen or the inferior alveolar canal.

DIFFERENTIAL DIAGNOSIS

Developmental—rather than pathologic—entities, such as anterior Stafne bone defect (ASBD) should be considered in cases of painless, asymptomatic, and unnoticed lesions without disconcerting features. However, unilocular, intrabony lesions in the tooth-bearing portions of the mandible may suggest odontogenic lesions, such as odontogenic keratocyst (OKC), adenomatoid odontogenic tumor (AOT), and glandular odontogenic cyst (GOC). Low-grade malignancies, such as central mucoepidermoid carcinoma (CMEC) are less likely but may manifest near the mandibular midline with well-demarcated borders. Finally, lack of other accompanying predisposing medical conditions could indicate an otherwise iatrogenic or idiopathic condition, such as idiopathic bone cavity (IBC). Metastatic or overtly aggressive lesions are unlikely in the absence of diffuse borders or neurologic and/or endodontic deficits. Other well-established entities, such as ameloblastoma or central giant cell granuloma, would be considered unusual without their characteristic expansion or multilocularity. Infectious or autoimmune pathoses are also unlikely if systemic symptoms, such as fevers and chills, or other extraoral manifestations are absent.

A Stafne bone defect first identified by its namesake investigator, is a nonpathologic defect of the bone cortex.[1] It was originally described as a unilateral and well-defined radiolucency of the mandible formed by pressure-induced loss of bone from a salivary gland.[2] The anterior variant (ASBD) is a rare presentation that often manifests as an ovoid radiolucency below or around tooth roots.[1] By definition, it resides anterior to the canines and does not exert notable pain, swelling, or loss of endodontic vitality—very much akin to its traditional counterpart.[1-3] Both Stafne bone defect and ASBD are classically diagnosed on the basis of incidental findings on

[a]Division of Oral and Maxillofacial Surgery, Alameda Health System, Highland Hospital, Oakland, CA, USA.
[b]University of the Pacific, Dugoni School of Dentistry, San Francisco, CA, USA.
[c]Division of Oral and Maxillofacial Surgery, College of Dental Medicine, Western University of Health Sciences, Pomona, CA, USA.
[d]Department of Oral and Maxillofacial Surgery, Loma Linda University Medical Center, Loma Linda, CA, USA.
Received for publication Aug 16, 2018; returned for revision Apr 28, 2019; accepted for publication May 7, 2019.

2212-4403/$-see front matter
https://doi.org/10.1016/j.oooo.2019.05.002

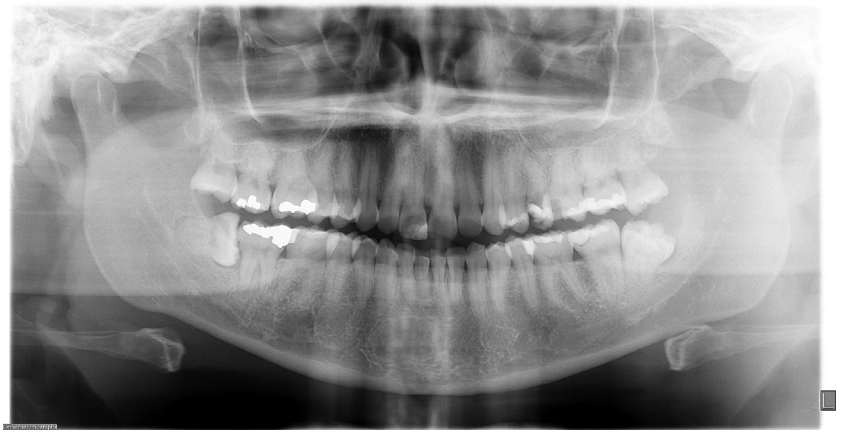

Fig. 1. Panoramic radiograph taken 1 year before presentation. The radiograph indicates no gross intrabony or dental pathology.

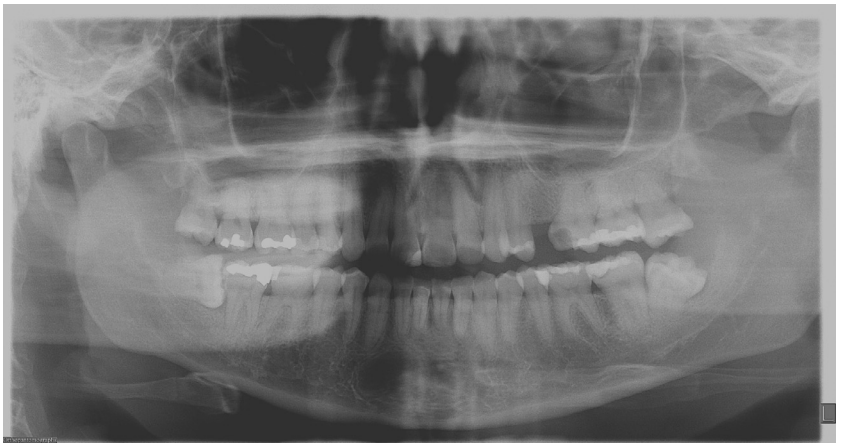

Fig. 2. A panoramic radiograph taken at the time of presentation. A circumscribed radiolucency is noted below the right mandibular incisors and canine (teeth #25, #26, and #27).

maxillofacial radiographs and may be further analyzed by using magnetic resonance imaging or sialography.[2] Many clinicians believe that they arise developmentally, and embryologic rests of salivary tissues have been found adherent to the mandible to lend credence to this theory.[1] Intriguingly, this discovery both supports and contradicts the current diagnosis of ASBD: Although a developmental entity could have been detected in a young adult, such lesions are unlikely to arise after an unremarkable panoramic radiograph taken only a year prior. Furthermore, the literature states that ASBD is much more commonly found in males and in middle-aged adults.[1,2] The diagnosis of ASBD in a young adult would be unusual, especially in the presence of the lingual cortex, which is virtually pathognomonic of the condition.[2]

In terms of odontogenic lesions, OKC is a well-known, relatively common entity that typically arises in young adults. Because of its frequency and its tendency to spread across the symphysis, OKC merits consideration in cases of cystic, intrabony lesions of the jaws approaching the midline. OKC most often presents with its characteristic unilocularity and distinct borders, and diagnosis may be incidental because obvious symptoms, such as paresthesia or overt swelling, may not be present.[4,5] Histologically, OKC is a benign but aggressive cyst of stratified squamous epithelium, and it grows as a result of increased mitotic activity of the basal cells in the dental lamina. Its proliferative potential usually causes an anterior-to-posterior direction of growth, allowing it to hollow the jaws in entirety but without expanding or perforating the cortices.[5] OKC is also strongly associated with nevoid basal cell carcinoma syndrome, suggesting a genetic component to this disorder.[4] Nonetheless, it appears to be more common among males and, possibly, those of Northern European descent.[6] It also develops with a marked predilection for the posterior mandible, especially in the ramus or the molar areas of the body. Its growth in the anterior mandible of a Hispanic female is relatively uncommon. A location significantly distant from appreciable odontogenic structures is also atypical.

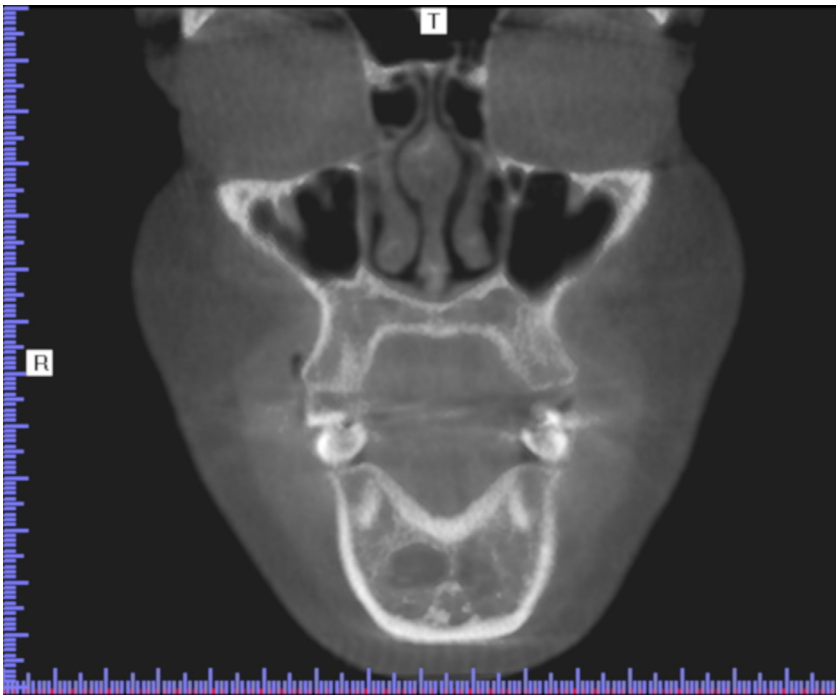

Fig. 3. Coronal view of the cone beam computed tomography (CBCT) image demonstrating a similarly well-circumscribed radiolucency observed in the panoramic radiograph.

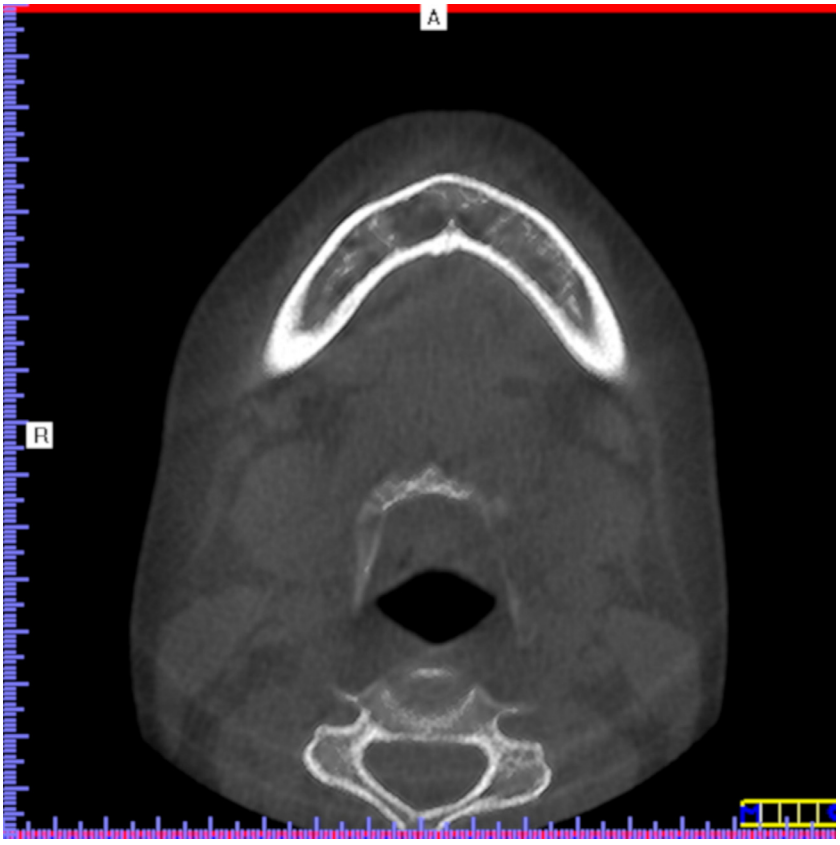

Fig. 4. Axial view of the cone beam computed tomography (CBCT) image suggesting no apparent interruptions to the buccal and lingual cortices surrounding the radiolucency.

Compared with the aggression that characterizes OKC, AOT is characterized by its modest expansion and round shape.[7,8] AOT was first documented in 1905 by Steensland, who described its hamartomatous origin (from the odontogenic epithelium), limited size, well-demarcated borders, and propensity for the anterior jaws.[9] Studies agree that both the follicular and extrafollicular variants of AOT demonstrate a predilection for females and the maxilla.[8-10] Although only the extrafollicular variants grow without notable physical attachments to odontogenic structures, both variants are marked by whorled and duct-like arrangements of epithelial cells along the foci of basophilic calcifications that are often present.[7,9,10] Both the follicular and extrafollicular lesions are likewise treated conservatively via enucleation.[7,9] Along with a preference for the maxilla, AOT fundamentally differs from the lesion in the present case in that it predominantly presents with radiopaque intralesional calcifications and during the first 2 decades of life.[7-9] AOT is also a common cause for the expansion, thinning, and discontinuity of the cortical plates, as well as displacement of nearby dentition, all of which were not seen on the radiographs in the present case.[7,9] Furthermore, an odontogenic entity, such as AOT, which occurs greater than 4 mm away from nearby dentition would be considered unusual.

Other than OKC and AOT, GOC is a rare lesion that is also of odontogenic origin. It was first described in 1987 by Padayachee and van Wyk, who noted its well-defined and unicystic appearance as well as its preference for the anterior mandible.[11-13] Although its exact pathophysiology has not yet been established, GOC was described as a slow-growing pathology marked by eosinophilic, cuboidal cyst lining; intraepithelial microcysts or duct-like spaces; and vacuolated cells with clear cytoplasms near or on basal layers, demonstrating a somewhat glandular appearance.[12,13] It is treated with conservative therapy consisting of enucleation, curettage, and, occasionally, peripheral osteotomy.[13] A subsequent survey by Fowler et al. found that the lesion commonly had no gender predilection.[12] Regardless of gender, a typical GOC patient population seems to consist of middle-aged individuals who report such symptoms as pain and swelling, which were not noted in our patient.[12,13]

CMEC represents a malignant process originating from salivary gland tissue. Its more common extraosseous presentation—simply denoted as mucoepidermoid carcinoma (MEC)—is frequently found predominantly in the parotid glands as a low-grade but potentially invasive tumor.[14,15] It represents an aberrant proliferation of the epidermoid and mucin-producing cells of the duct linings, with possible bleeding, paresthesia, or motor neuron deficits in nearby structures.[14] CMEC has similar invasive potential as MEC but usually mimics bony mandibular cysts with uniloculation or multiloculation and cortical preservation.[14] Previous studies have reported that CMEC arises from (1) entrapped minor mucous glands that transform, (2) embryonic remnants of the submandibular/sublingual glands that become encased in bone, or (3) neoplastic glandular tissue that invade from the periosseous soft tissue linings.[15] Regardless of origins, treatment of all MECs requires wide surgical resection, followed by possible radiotherapy and neck dissection in cases of advanced presentation.[14,15] Microscopy also reveals infiltrative processes, with proliferation of nests, islands, and cystic structures composed of epidermoid, mucous, and intermediate cells.[15] Although CMEC usually occurs in women in their third to sixth decades of life, its rarity and predilection for the posterior jaws made it an unlikely diagnosis in the present case.[14] There was a likelihood of a unilocular presentation of CMEC, but it was not significant.[14,15]

Unlike the soft tissue proliferations (OKC, GOC, and CMEC), IBC is an intraosseous cavity that was previously thought to be associated with trauma without a clearly defined cause. It was first documented in 1929 by Lucas as a radiographically well-defined, unilocular, empty cavity of the mandible, with scant fibrous tissue, normal bone, and occasional clear fluid.[16,17] An effective treatment strategy involves inducing intracavity bleeding via curettage, indicating that the entity or injury simply lacks the reparative factors present in ordinary blood.[16] Approximately 30% of all IBCs present in the anterior mandible, with a small likelihood of cortical enlargement. In most cases, it is detected incidentally because it has no notable symptoms.[16,17] Posterior mandible manifestations and a predilection for teenaged males are the predominant features of IBC.[16,17] IBC is also characterized by its propensity to form scalloped superior borders around tooth roots.[16]

DIAGNOSIS AND MANAGEMENT

An aspirational biopsy was first performed, and it did not yield notable fluids or cell aspirates. The lack of fluids indicated that the entity was not cystic in nature. During a subsequent excisional biopsy, a bony window revealed a doughy, yellow-white soft tissue mass, which measured approximately $30 \times 20 \times 10$ mm and appeared distinct from the surrounding bony structures. The lesion was first detached from its bony cavity by using a broad curette and then removed in a single piece by using blunt forceps. Manipulation of the mass did not yield any blood or fluids. The cavity was then curetted thoroughly, and the intact pathologic specimen was submitted for pathologic analysis. The specimen floated within the container of formalin, suggesting a lipomatous character.

Histopathology was performed with hematoxylin and eosin staining. Microscopy demonstrated a mass of benign, mature adipose tissue intermixed with lamellar bone (Figure 5). There were no signs of lipoblasts or hyperchromatic stromal cells indicative of liposarcoma

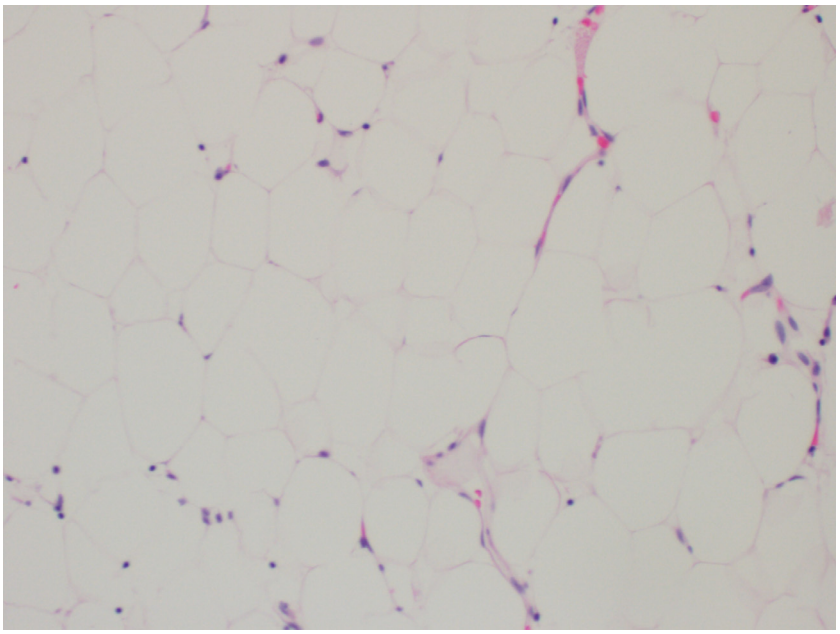

Fig. 5. A × 200 view of the intermixed adipose and bone tissue without signs of hematopoietic, fibrous, or vascular entities. *A high-resolution version of this slide for use with the Virtual Microscope is available as eSlide: VM05600.*

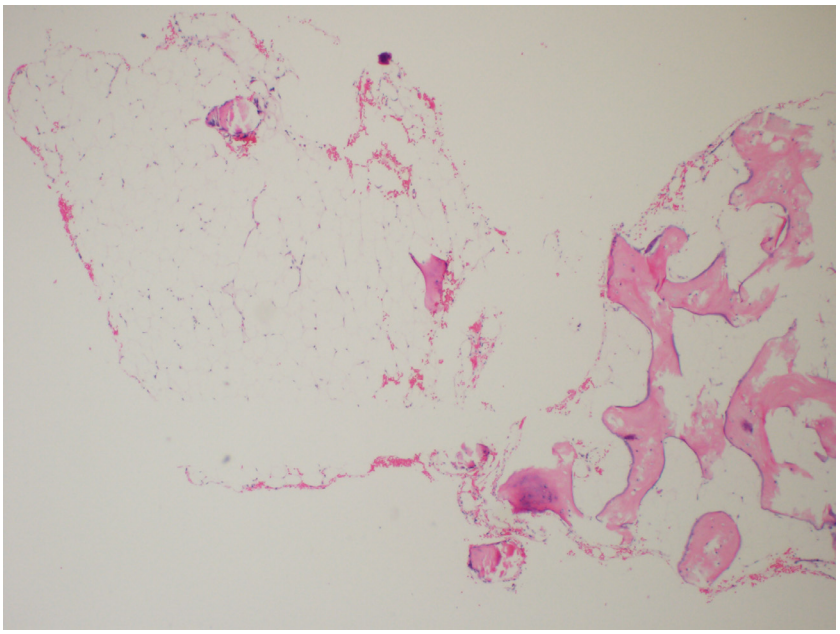

Fig. 6. A × 200 view of mature adipocytes without signs of lipoblasts, hyperchromatic stromal cells, increased mitoses, nuclear enlargement, or inflammatory infiltrations. *A high-resolution version of this slide for use with the Virtual Microscope is available as eSlide: VM05596.*

(Figure 6). The specimen also lacked mitotic figures, increased nuclear-to-cytoplasmic ratio, or lymphocytic and multinucleated giant cell infiltrations.

On the basis of the clinical and radiographic findings, a diagnosis of mandibular intraosseous lipoma (MIL) was made. A further referral was made, and the sample was sent to an oral and maxillofacial pathologist to confirm these findings. No additional treatments were recommended or indicated according to the established guidelines in the literature. Only periodic clinical and radiographic monitoring was required.

DISCUSSION

Intraosseous lipoma (IL) is an uncommon, benign lesion of bones, making up approximately 0.1% of all primary osseous tumors. It is typically discovered in

the calcaneous long bones and has a predilection for males in their fourth decade of life.[18] Presentation within the mandible remains particularly rare, with no more than 20 cases of MIL documented within the English language literature.[18-21] Interestingly, reviews of MIL cases have found a predilection for MIL to occur in females and those in their fifth and sixth decades of life.[19,22] It is possible that the unique intraoral and mandibular environments require a different set of circumstances for the growth of an otherwise similar lipomatous lesion.

MIL has been documented to cause swelling, pain, and paresthesia, depending on its location and size, but could exist asymptomatically for several years.[22] Histologically, both MIL and IL are characterized by sheets of mature adipocytes without accompanying lipoblasts, hematopoietic elements, or atypia.[18] The source of its cells of adipocytic lineage are presumed to be fatty marrow, the periosteum, or adjacent soft tissue.[22] A study of Brazilian patients showed chromosomal aberrations in most of its MIL specimens, whereas long bone IL has been associated more with trauma, ischemic infarction, and inflammation.[19] Although the exact mechanism is difficult to ascertain, most studies agree that IL, in general, is a true benign tumor of the medullary adipose tissue.[20]

Classification of MIL and IL cases has been understandably difficult. In many cases, MILs appear to vary in their degree of encapsulation and location in relation to the bony cortex, with masses described as intramedullary, intracortical, parosteal, and subperiosteal.[18] Milgram et al. have also proposed a numerical staging scheme based on histology, with stage 1 showing normal adipocytes, stage 2 showing partial necrosis, and stage 3 demonstrating complete secondary necrosis and dystrophic calcification.[23] Thus, we could characterize the lesion in the current case as stage 1 intramedullary MIL. Regardless of classification, the overall prognosis for MIL remains excellent, with enucleation and curettage often resulting in complete elimination of disease. There are also reports of incomplete enucleations leading to complete disease regression without future recurrences.[19]

As the overall number of MIL cases is small, establishing generalizations regarding the nature of MIL is difficult. However, in the current case, the patient presented with several features that were notably different from those in previously documented cases. First, the medial border of this lesion was found to be closely approximating the symphysis compared with the majority of reports describing a posterior location.[22,24] Second, the current patient was a young female in her early 40s, that is, at the lower end of the spectrum of known ages of presentation.[19,21,22]

The nonaggressive behavior of MIL indicates that in most cases, improvement will be seen even after conservative treatment. However, the extremely limited number of reports so far necessitates detailed documentation of all new cases to better ascertain the nature and tendencies of MIL.

CONCLUSIONS

Indeed, the findings in our case lend additional credence to the fact that MIL may manifest asymptomatically and that it could be seen as a homogeneous, unilocular mass on standard dental radiographs. And although some of its characteristics have been previously described, we found a relatively novel presentation of MIL in the anterior mandible of a young, healthy female patient.

ACKNOWLEDGMENT

We wish to thank Lee J. Slater, DDS, MS (Staff Pathologist, Scripps Oral Pathology Service, San Diego, CA), for his assistance with histopathologic evaluation and with the preparation of the manuscript.

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Reprint requests:

Dr. Jeffrey A. Elo
Division of Oral and Maxillofacial Surgery
College of Dental Medicine
Western University of Health Sciences
795 E. Second St.
3rd Floor
Pomona
CA 91766
(909) 706-3910
fax: (909) 469-8650
USA.
jelo@WesternU.edu