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Massive, mixed, cystic lesion of the mandibular midline

Hiba Qari, BDS, MS,[a] Garrett Blundell, DDS,[b] Nagi Demian, DDS, MD,[b] Michael Covinsky, MD, PhD,[c] and Jerry E. Bouquot, DDS, MSD, FICD[a,d]
Baylor College of Dentistry, Dallas, TX, USA; University of Texas, Houston, TX, USA
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CLINICAL PRESENTATION

A 37-year-old white man presented to the emergency clinic of a teaching hospital complaining of a "swelling" of his chin of 5 months' duration. While he was playing with his daughter, she accidently hit his chin, resulting in increasing size, numbness, and pain during eating. The patient had no significant medical history, but he had smoked cigarettes (1 pack per day) since his teenage years. Clinically, the chin mass was large (7 × 6 × 4 cm), with a slight asymmetry of the symphysis region. No skin changes, fever, or purulent discharge were observed.

Intraorally, there were multiple carious teeth, the mandibular left canine was missing, and the remaining teeth from right second mandibular premolar to left second premolar exhibited class III mobility; many were haphazardly displaced. There was slight pain during percussion of most mandibular teeth. The lesion appeared to be intraosseous, with a considerably expanded cortex that was nontender and pliable on both labial and lingual surfaces.

A panoramic radiograph showed a well-circumscribed radiolucency extending from the right first mandibular molar to the left first molar, with an impacted left canine displaced to the inferior border. Lateral oblique radiographs helped to further characterize the lesion (Figure 1). The lesion had caused displacement and considerable external root resorption of overlying teeth, as well as thinning, expansion, and slight scalloping of the inferior mandibular cortex. There was a thin sclerotic rimming on the left.

Conventional computed tomography imaging (soft tissue window, with contrast) confirmed the well-circumscribed nature of the lesion and also showed irregularly dense regions of mild opacity, consistent with soft tissue proliferations, along the lesional periphery and occasionally more centrally (Figure 2); some of these regions had enough density to suggest at least mild calcification. The bulk of the lesion was represented by a mildly and diffusely gray haze, suggesting a fluid content. There were several small, more radiopaque densities of the posterior portions, and the overlying cortex was very thin, with occasional perforations not evident on other radiographs. The lesion considerably expanded toward the labial, but also showed expansion of the lingual aspect of the mandible.

DIFFERENTIAL DIAGNOSIS

The massive size of this lesion, with its multiple cortical perforations and associated paresthesia, suggested an aggressive intraosseous neoplasm. However, the well-demarcated borders, cortical expansion, sclerotic rimming, and resorbed tooth roots suggested a benign neoplasm or cyst and almost certainly ruled out primary malignancy or metastatic disease. It was thought that the perforations might be explained by palpation trauma in such an unusually thinned cortex, or that perhaps the "perforated" areas were merely regions where the cortex was so thin that it could not be detected by computed tomography technology.

The paresthesia would be more difficult to accept for a benign entity if the paresthesia were not bilateral, suggesting a common pressure or ischemic phenomenon from tumor expansion.

Three additional and significant features of the lesion provided important clues to its identity: (1) it was not a solely pericoronal lesion, despite its association with an impacted tooth; (2) it was slightly multilocular, as suggested by scalloping of the inferior border; and (3) although basically a cystic radiolucency, it contained scattered radiopacities. The fact that it extended beyond the cementoenamel junction (see Figure 1, C) broadened our differential diagnosis and ruled out at least one expansile radiolucency, the dentigerous cyst. Additionally, the multilocular appearance suggested a number of classical lesions, such as ameloblastoma, odontogenic keratocyst (keratocystic odontogenic tumor), glandular odontogenic cyst, odontogenic myxoma, ameloblastic fibroma, central giant cell granuloma, aneurysmal bone cyst, intraosseous mucoepidermoid carcinoma, and central hemangioma.[1] All can be associated with pronounced cortical expansion, but only one, the central giant cell granuloma, is capable of producing

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[a]Department of Diagnostic Sciences, Baylor College of Dentistry.
[b]Department of Oral & Maxillofacial Surgery, University of Texas School of Dentistry at Houston.
[c]Department of Pathology & Laboratory Medicine, University of Texas Medical School at Houston.
[d]Department of Diagnostic & Biomedical Sciences, University of Texas School of Dentistry at Houston.
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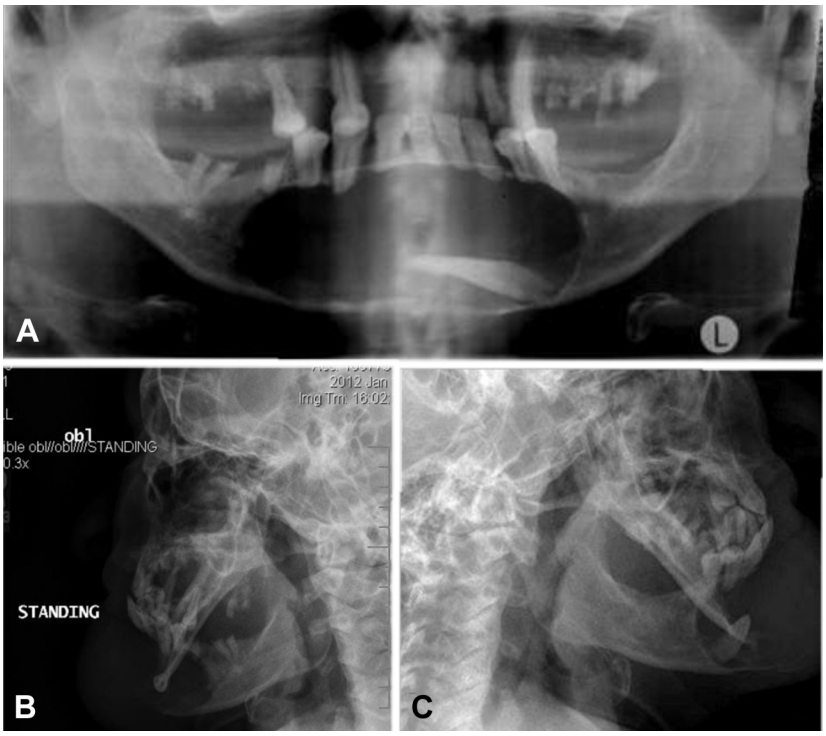

Fig. 1. **A,** Massive, well-demarcated, unilocular radiolucency of the anterior mandible associated with an impacted cuspid. It demonstrates severe root resorption and tooth displacement. **B, C,** Lateral oblique jaw radiographs help orient the position of the impacted tooth.

radiopacities, representing exuberant new bone formation at the lesional periphery. All the others, for that reason, could be excluded from the differential diagnosis.

It appeared that the mixed nature of the radiographic presentation was the most significant diagnostic sign, one that suggested a number of other benign odontogenic and osseous lesions, in addition to the giant cell granuloma previously mentioned. Odontogenic, mixed-radiopacity candidates capable of expanding the cortex and becoming multilocular include[1] odontoma, ameloblastic fibro-odontoma, odontoameloblastoma, calcifying odontogenic cyst (Gorlin cyst), calcifying epithelial odontogenic tumor (Pindborg tumor), adenomatoid odontogenic tumor/cyst (AOT), and central odontogenic fibroma (World Health Organization [WHO] type).

The odontoma, odontoameloblastoma, and ameloblastic fibro-odontoma, however, could reasonably be ruled out, because the opacities in the present lesion were not as dense as the opacities of those entities.

The Gorlin cyst, also referred to as the calcifying cystic odontogenic tumor, especially when it is more solid than cystic, is most frequently found in the anterior region, as was the present lesion.[1,2] Its mean age at diagnosis, 33 years, is very close to the present patient's age, although the more aggressive, solid variants tend to occur in older individuals. It is almost always unilocular, but occasional multilocular cases have been seen. Also, it is adjacent to an impacted tooth, usually a cuspid, about a third of the time, and it frequently causes tooth displacement and root resorption. The radiopacities of the Gorlin cyst are not always present but may be less dense and irregular, as in the present case, or may be denser, even tooth-like. This cystic lesion can become huge, with reported cases reaching up to 12 cm in diameter. It appeared to be a good diagnostic candidate for the present case.

The Pindborg tumor can show massive expansion as well. It is frequently associated with an impacted tooth and has been most often found in persons 30 to 50 years of age, with no gender predilection, so it was acceptable as a differential diagnosis for the present case.[1,3,4] Additionally, two-thirds of all cases are mandibular, although almost all have been posterior, not anterior. The tumor can be unilocular, but many multilocular examples have been reported, especially when occurring in the mandible. Its scattered calcified structures typically remain small and are of variable density. Overall, this diagnosis was a very viable candidate for the differential diagnosis of the present case. It should be mentioned here, also, that there are malignant or borderline-malignant variants of the Pindborg tumor, and the cortical perforations and paresthesia might be explained by this potential variant.