A 22-year-old woman presented with discomfort of the left posterior maxilla that had been present for several days. The patient’s medical history was noncontributory. On clinical examination her left maxillary first and second molars were free of caries and dental restorations. Clinically, no third molar was seen. The maxillary ridge posterior to the molars was normal. There was no pain on percussion of the molars or on palpation of the area. Examination of the temporomandibular joint was within normal limits. A panoramic radiograph revealed that the maxillary and mandibular left third molars were impacted. The left maxillary third molar showed a well-defined radiopacity around its apex that was round, uniform in appearance, and surrounded by a narrow radiolucent rim (Fig. 1). It measured approximately 1.1 cm in diameter.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis of a localized, periapical radiopacity includes true periapical lesions, such as cementoblastoma, hypercementosis, periapical/focal cemento-osseous dysplasia, and focal sclerosing osteomyelitis, as well as lesions that may coincidentally be found near a tooth apex, such as odontoma, cemento-ossifying fibroma, osteoma, and osteoblastoma/osteoid osteoma. Underlying the need for the formulation of a thorough differential diagnosis is the observation that the biologic behavior of these lesions varies from totally innocuous to aggressive, and the recommended treatment from simple radiographic follow-up to surgical excision.

A cementoblastoma (“true cementoma”) is a rare tumor of neoplastic cementoblasts.1 Radiographic examination usually shows a well-defined, circumscribed radiopacity confluent with the root or roots of the tooth, surrounded by a thin radiolucent halo. Root resorption, loss of root outline, and obliteration of the periodontal ligament space may also be seen. Based on a review by Brannon et al.2 of 44 of their own cases and an additional 74 cases identified in the literature, the mean age at presentation was 21.3 years, with a slight male predilection (1.4:1). Both jaws were affected, although 79.5% of the cases were located in the mandible, most commonly associated with the first mandibular permanent molar. Most cases presented with cortical expansion and pain. Cementoblastoma is considered to represent a benign neoplasm with locally aggressive biologic behavior and a high recurrence rate (21.7%-37.1%). Surgical excision of the lesion and the affected tooth, followed by curettage or peripheral osteotomy is recommended. At least 3 cases of cementoblastoma associated with unerupted or impacted teeth have been reported.2

Hypercementosis, representing excessive deposition of cementum around the apex of a tooth, may present as a well-defined radiopaque mass fused with the root of a vital tooth, resulting in a “club-shaped” appearance.3 This excess cementum may be surrounded by a thin

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radiolucent halo of uniform width, representing the normal periodontal ligament space. This condition has been associated with various local stimuli (trauma, altered function, inflammation) and systemic factors (Paget’s disease, hyperthyroidism), but in most cases represents an idiopathic, age-related phenomenon.3,4 It is most common in the premolar area and, as it is asymptomatic, is usually found during routine radiographic examination. Surgical excision is performed for diagnostic purposes only in atypical cases. The lack of opposing teeth is a predisposing factor for hypercementosis1 and although no cases comparable to the present one were found in a review of the literature, marked hypercementosis could not be entirely excluded from the differential diagnosis.

A low-grade inflammatory stimulus, such as chronic periapical inflammation or malocclusion, may be the causative factor of focal sclerosing osteomyelitis (condensing osteitis, periapical osteosclerosis). When no apparent cause can be identified, the term idiopathic osteosclerosis is applied.2 Condensing osteitis and idiopathic osteosclerosis are the most common periapical radiopacities found in adults and are typically discovered during routine radiographic examination. They commonly present as a uniform periapical radiopacity around a mandibular molar tooth, although they can be noted anywhere in the jaws. No peripheral radiolucent rim is noted. The periodontal ligament space and lamina dura can usually be identified around neighboring teeth.

Cemento-osseous dysplasias are relatively common fibro-osseous lesions of the tooth-bearing areas of the jaws.1 They are generally asymptomatic and usually discovered on routine radiographic examination.5,6 Approximately 70% of cases are found in close proximity to the apices of vital teeth (periapical cemento-osseous dysplasia), commonly the mandibular anterior teeth of middle-aged women.6 They start as periapical radiolucentencies that may “mature” over time into circumscribed radiopacities surrounded by a radiolucent rim.6 Periodic radiographic follow-up is the treatment of choice.5,6 While focal cemento-osseous dysplasia could be included in the differential diagnosis of the present lesion, the association of an impacted tooth with cemento-osseous dysplasia has not been reported.

Odontomas are composed of dentin and enamel in amorphous conglomerations (complex odontomas) or in rudimentary tooth-like structures (compound odontomas).1 Odontogenic epithelium and mesenchyme may also be found, depending on the stage of development. Odontomas are usually discovered incidentally in children or young adults during routine radiographic examination or during investigation of a missing or displaced tooth. Complex odontomas are more common in the posterior mandible and present as a well-defined, uniform radiopacity surrounded by a radiolucent rim that represents a fibrous capsule. Variations in the degree of radiopacity due to the presence of various dental tissues may facilitate differential diagnosis from other radiopacities.7 The presence of multiple tooth-like structures of various size and shape is diagnostic of compound odontoma. These are more common in the anterior maxilla.1 Simple excision is curative.1 There is at least 1 report of a complex odontoma in the periapical and interadicular area of a second primary mandibular molar with associated root resorption.7 Superimposition of an odontoma over the root of an impacted tooth was a definite consideration in this case.

Ossifying fibroma (cemento-ossifying fibroma) is a rare, benign bone neoplasm that is more frequently found in the posterior mandible.1 The radiographic appearance of this lesion varies from a unilocular radi-
olucency to, rarely, a radiopaque mass surrounded by a well-defined, radiolucent rim.\(^1\) Presentation restricted to a periapical location is extremely unusual.\(^6\) Although this tumor usually displaces the roots of adjacent teeth, on occasion it may cause root resorption resulting in union with the associated teeth.\(^1\) Surgical removal by curettage or enucleation is recommended.

The osteoma is a benign tumor of the jaws, most commonly encountered in young patients, that usually involves the body of the mandible or the condyle. Multiple osteomas may be a component of Gardner’s syndrome.\(^1\) An endosteal osteoma presents as a small, asymptomatic radiopacity without a radiolucent rim. Only periodic radiographic examination is required. The osteoblastoma/osteoid osteoma typically is larger and may be associated with pain.\(^1\) This benign bone neoplasm typically involves the posterior mandible and shows a predilection for young males. Recurrence is rare after conservative excision or curettage.

**MANAGEMENT AND DIAGNOSIS**

Under local anesthesia, a mucoperiosteal flap was raised posterior to the maxillary left second molar. The cortical bone was removed, exposing the impacted maxillary third molar. A globular mass protruding from the tooth root was seen. The third molar tooth and attached globular mass were easily shelled out en block and the surgical flap was repositioned and sutured. Healing was uneventful.

Macroskopically, the extracted tooth had the coronal morphology of a molar with a single root. A hard, globular mass, measuring 1.2 cm in diameter, was seen at the apex of the tooth. The globular mass consisted of an extension of the radicular dentin that encircled a mass of hard tissue (Fig. 2). Microscopic examination showed that the dentin surrounding the mass was primarily mature dentin with well-oriented tubules continuous with the radicular dentin (Fig. 3). Also noted were enlarged tubules, and circular or oval cavities that contained connective tissue. The dentin was lined by enamel and the inner mass consisted of enamel matrix, foci of osteoid/cementoid material, and connective tissue. A communication of the dilation with the outer root surface could not be located with certainty, although a small brown line suggestive of such a communication was present at the apical end of the globular mass (Fig. 2). A discontinuity at the apical end was a constant feature in all microscopic slides examined.

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**Fig. 2.** Macroscopic examination shows a dilation of the radicular dentin that contains a mass of hard tissues. The arrow highlights the probable site of communication of the dilation with the outer root surface.

**Fig. 3.** The dilation of the radicular dentin (D) is lined by enamel (arrows) and contains mostly enamel matrix (E). P indicates pulpal cavity (hematoxylin and eosin stain, inverted digital picture, original magnification \(\times1\)).
DISCUSSION

Dens invaginatus (dens in dente, “tooth within a tooth,” dilated composite odontome) is a developmental dental abnormality presenting as a deep, enamel-lined invagination in the crown (“coronal dens invaginatus”) or root (“radicular dens invaginatus”). The diagnosis was consistent with radicular dens invaginatus.

The clinical features of previously reported cases of radicular dens invaginatus and the present case are summarized in Table I. Seven of the 8 patients were females, mostly younger. The most commonly affected teeth were incisors (3 cases) and third molars. The case reported by Bhatt and Dholakia was unusual in that the enamel-lined invagination contained a smaller enamel invagination, forming a “radicular double dens in dente.” No cases were reported in association with an impacted tooth. Three patients exhibited pain from abscess formation or pericoronitis. Pulpal inflammation has been attributed to incomplete lining of the radicular cavity by enamel and its extension into the usually hypoplastic pulp, permitting entrance of bacteria from the periapical or periodontal space.

Radicular dens invaginatus has been attributed to the proliferation and ingrowth of Hertwig’s epithelial root sheath (HERS) into the dental papilla, followed by dental follicle connective tissue. Subsequently, HERS cells differentiate into ameloblasts, while dental follicle mesenchymal tissue forms cementum and bone. During normal odontogenesis, most HERS cells are lost through apoptosis after deposition of the first layer of radicular dentin. Subsequently, HERS develops fenestrations and dissolves, leaving small groups of cells in the periodontal ligament, known as rests of Malassez. In order for HERS cells to proliferate and survive in contact with radicular dentin, a deficiency in the cell death mechanism of those cells could be anticipated. Hamamoto et al. showed that when cells of the HERS-derived epithelial islands in the apical periodontium of rats come in contact with dentin, they acquire the potential to differentiate into ameloblasts and produce amelogenin. This is in contrast to the reported inability of normal HERS cells to differentiate into ameloblasts, but may explain “the formation of ectopic enamel in radicular complex odontoma or odontoma with adhesion to the root of tooth.” Recent studies have shown that HERS regulates root formation by acting as a signaling analogous to the enamel knot of the crown. Deregluation of the HERS’ radicular signaling center is a likely explanation for the formation of radicular dens invaginatus.

Rushton suggested that dens invaginatus and “complex composite odontome” share a common pathogenetic process, and used the term “dilated composite odontoma” for the former. Rushton proposed that what differentiates them is that in dens invaginatus the dis-
turbance occurs later in odontogenesis, thus the tooth germ follows a largely normal differentiation pattern, thereby forming a recognizable tooth. According to this view, radicular dens invaginatus can be considered a “mature” odontoma, the distinction from “conventional” odontoma representing a matter of degree.

The differential diagnosis in the present case also included a complex odontoma fused with the tooth root. According to our review of the English-language literature, only 1 case of a periapical complex odontoma fused with a tooth has been reported. The lesion was attached to the interradicular region of a retained second primary molar and was composed mainly of cementum-like tissue with areas of enamel and dentinal tissue. It had completely resorbed the mesial root, and was separated from the adjacent tooth by a fibrous capsule. In our case, the globular mass consisted of a dilation of the radicular dentin that was lined by enamel, and contained mostly enamel matrix, cementoid/osteoid material, but not dentin. Also, in contrast to an odontoma, it was not surrounded by a fibrous capsule. Although a communication of the outer tooth root with the dilation could not be documented in our case, it may have been overlooked during the sectioning process. Thus, we suggest that our case is consistent with a radicular dens invaginatus, and is quite similar to cases 17 and 18 reported by Rushton.

REFERENCES

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