Atypical hard tissue formation around multiple teeth

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Over the preceding 4 months, a 36-year-old man had noticed an irritation of the gingiva in the upper left premolar and molar region with exposure of root surfaces. He did not complain of pain. Other than a history of smoking (20 cigarettes per day, 20 cumulative pack-years), the patient did not present with any medical problems and was not taking any medications. The patient, who was a farmer, reported a trauma which had taken place 15 years earlier, in which a cow horn had perforated his left cheek. No dental trauma had been noted and no radiographs had been taken at that time.

On clinical investigation, a flattening of the left cheek and a scar in the overlying skin were visible. No intraoral swelling or expansion was present. On the buccal side of the upper left first molar and in the interdental space next to the second premolar, extensive gingival recession with exposure of a brownish irregular hard dental structure was conspicuous (Fig. 1). The hard structure seemed to be fused to the roots. Periodontal pocket probing in the upper left jaw gave results of ≤4 mm, except buccal pocket probing of the second premolar (6 mm), buccal pocket probing of the second molar (5 mm), and distal pocket probing of the second molar (7 mm). The maxillary left third molar was partially erupted. With the exception of the second premolar, all teeth in the upper left maxilla reacted positively to cold. A periapical radiograph, which had been taken by the referring private dentist, exhibited diffuse radiopaque structures overlapping the roots of the molars and premolars (Fig. 2). For further evaluation of the unusual structure, cone-beam computerized tomography (CBCT; volume size 6 × 6 cm, 80 kV, 5 mA, 6 mGy; 3D Accuitomo XYZ Slice View Tomograph, J. Morita Mfg. Co., Tokyo, Japan) was performed. Serial imaging in sagittal, coronal, and axial sections demonstrated extensive radiopacity encompassing the upper left first and second premolars, as well as the first and second molars, with no visible periodontal space (Fig. 3). Apical to the radiopaque mass and the mentioned teeth, as well as distal to the root of the second upper molar, a radiolucent zone of up to 10 mm was observed, with a continuous bony demarcation toward the left maxillary sinus. In the left maxillary sinus, basal thickening of up to 12 mm of the sinus mucosa was present.

DIFFERENTIAL DIAGNOSIS

Based on the clinical examination and the periapical and CBCT imaging, the lesion was presumed to be of cemental origin. Therefore, the differential diagnosis included cementoblastoma, ossifying fibroma, osseous dysplasias, and hypercementosis. Although it was less likely, we also considered the possibility of an osteosarcoma.
Cementoblastoma, characterized by the formation of cementum-like tissue in connection with a root of a tooth, is classified as a benign ectomesenchymal odontogenic tumor. It is a rare tumor, with <100 cases documented in the English-language literature. Cementoblastoma is most often located in the mandible, typically related to a permanent first molar. Premolars are the second most often involved teeth, and single cases have been described in association with deciduous teeth, incisors, or unerupted teeth. It is a neoplasm occurring typically in younger patients, with a peak between the ages of 20 and 30 years. The male-to-female ratio is about 1.4:1. Most cementoblastomas involve 1 tooth. However, in the literature there are also cases describing the involvement of multiple teeth. Overall, 80% of cementoblastomas are localized in the mandible, but interestingly, when multiple teeth were involved, cementoblastomas occurred more often in the maxilla. Clinical signs can include swelling and/or pain, but asymptomatic cases have also been described. The radiographic appearance in the early stage is characterized by osteolysis; this is gradually replaced by radiopaque material, leaving just a small radiolucent rim in later stages. The periodontal space disappears, and root resorption is a common finding. In the present case, the patient was older than the peak age reported for cementoblastomas. He did not show the typical symptoms of pain or cortical expansion of cementoblastoma, but on the buccal side a hard brownish structure and gingival retraction were present. The extensive radiopacity, as well as the radiolucent apical border on the CBCT images, could indicate a late-stage cementoblastoma. Because multiple teeth were involved, the maxillary location was not atypical. As is characteristic of cementoblastomas, the periodontal space was not visible, but the root shape could clearly be recognized and did not show any signs of resorption.

Cemento-osseous dysplasias encompass a spectrum of conditions characterized by dysplastic bone and cementum formation. Three variants have been described: periapical cemento-osseous dysplasia, florid cemento-osseous dysplasia, and focal cemento-osseous dysplasia. In the 2005 World Health Organization (WHO) Classification of Tumors of the Head and Neck, the term “cemento” was eliminated, because these lesions are characterized by replacement of normal bone. The various forms are indistinguishable histologically, but have different clinical appearances. Periapical osseous dysplasia is most often seen in middle-aged black women, and is localized exclusively to the anterior mandible. Florid osseous dysplasia also affects mostly middle-aged black women. It is a multifocal condition, usually presenting bilaterally in the mandible. The only variant of osseous dysplasia that we considered in the present case was focal osseous dysplasia, which is a localized lesion in a site other than the
anterior mandible. Although the mandibular posterior site is most often affected, the maxilla is also affected in one-fourth of cases. Osseous dysplasia appears most often between the third and sixth decades of life, with a peak frequency in the fourth and fifth decades. Up to 90% of patients are female. Radiologically, osseous dysplasia has various appearances, including radiolucent presentation with or without a sclerotic rim, lesions with mixed radiolucency and radiopacity, and lesions with pure radiopaque areas. Lesions are often noted incidentally during radiographic examination, because they are clinically asymptomatic. Although less common in men and in the maxilla, based on the clinical and radiologic considerations of the present case, focal osseous dysplasia could not be ruled out.

The ossifying fibroma is a well demarcated to encapsulated benign neoplasm consisting of fibrous tissue containing variable amounts of mineralized material. The mineralized component may consist of woven bone, lamellar bone, and acellular to poorly cellular deposits thought to be cementum. However, cementum is defined as a mineralized material only covering the surface of roots of teeth, and usually not present in other locations. Since the WHO classification of 2005, the term “ossifying fibroma” has been used for this bone-related lesion. These lesions occur in the tooth-bearing areas of the jaw, with up to 80% occurring in the mandible, most often in the premolar-molar region. A significant number of ossifying fibromas also involve the maxillary molar region. The mean age at first presentation of ossifying fibroma is 31 years, and most cases occur in the third to fourth decades of life, which would correlate with the age of the present patient, although the preferred gender is female. Typical signs of ossifying fibroma include swelling or jaw expansion, but one-third to one-half of patients are asymptomatic on initial presentation. Radiographically, the tumor is a well defined unilocular or, in rarer cases, multilocular lesion with or without a sclerotic border. The mixed radiopaque and radiolucent lesions predominate, but the lesions can also present as predominantly radiopaque or radiolucent. Involved teeth are typically displaced. Associated roots can be resorbed. In the CBCT sections of the present patient, the pathology was a well demarcated radiopacity in direct contact with the roots of the teeth, without any evident root displacement or resorption.

Fig. 3. Cone-beam computerized tomography of the left maxilla, displaying the premolar and molar region as well as the maxillary sinus. A, Sagittal view. B, Axial view. C, Coronal view in the region of the mesiobuccal and palatal roots of the first molar. D, Coronal view in the region of the mesiobuccal root of the first molar. E, Coronal view in the region of the distobuccal and palatal root of the first molar. F, Coronal view in the region of the second molar.
Osteosarcoma of the jaws is a rare malignant neoplasm occurring mostly in the third to fourth decades of life, though also occurring in teenagers.\textsuperscript{5,13} Because the clinical signs and symptoms are similar to various infectious or benign neoplastic diseases, a significant period often elapses before the correct diagnosis is established. The most common symptoms are swelling, expansion of bone, pain, and paresthesia. Mobile teeth or an incidental radiographic finding of a radiopacity or a widened periodontal ligament are more subtle signs of this neoplasm.\textsuperscript{5} Larger case series have shown a mandibular predominance.\textsuperscript{14} However, some smaller series described more tumors in the maxilla than in the mandible.\textsuperscript{13} Maxillary tumors seem to predominate in the posterior part of the alveolar process and the antrum.\textsuperscript{14} Radiologic features of osteosarcoma can be difficult to interpret, contributing to a delay in diagnosis. The tumors are radiolucent, radiopaque, or of mixed density. Typical radiologic signs include a sun-ray appearance, which is not often seen in smaller lesions, and a widening of the periodontal ligament space of involved teeth. Interpretation of these signs is also dependent on the type of radiograph examined.\textsuperscript{15} The present patient did not have any typical symptoms or aggressive tumor growth, but because the features of an osteosarcoma can vary, it could not absolutely be excluded based on the radiologic interpretation of CBCT imaging alone.

Hypercementosis is a nonneoplastic condition of excessive cementum deposition, and it may be associated with root ankylosis when cementum is in direct contact with the alveolar bone. Hypercementosis is symptomless, and tooth sensitivity is mostly unaffected. Hypercementosis can lead to fusion of the roots of adjacent teeth.\textsuperscript{16} Thickening of the cementum is widely accepted as an age-related phenomenon.\textsuperscript{17} Other etiologic factors, such as occlusal forces, chronic periapical inflammatory processes, unerupted teeth, and systemic factors, have been reported.\textsuperscript{16,18-20} On radiographic examination, a bulb-like enlargement of the roots is seen, surrounded by normal periodontal space and a radiopaque lamina dura.\textsuperscript{20} On the CBCT images of the present patient, a normal periodontal ligament space was found mesially and apically to the root of the first premolar, whereas an osteolysis of up to 10 mm was seen apically of the second premolar and both molars as well as distally of the second molar. In addition to the radiopacities between the teeth, apical nodular root enlargements could be recognized on the axial view of the second premolar and both molars.
To establish a definitive diagnosis, the decision was made to remove both molar teeth, as well as the peri-apical soft tissue, leaving the premolars in situ. A mucoperiosteal flap was raised in the upper left quadrant under local anesthesia and aseptic conditions. A brownish-white hard substance with an irregular surface could be observed around and in between the roots of both premolars and molars (Fig. 4). The first molar was separated from the second premolar using ultrasound osteotomy and extraction tips (Piezosurgery; Mectron Medical Technology, Carasco, Italy). The 2 maxillary left molars were immersed in 4% buffered formalin. The apical soft tissue was curetted from the underlying bone and submitted separately. The base of the alveolar defect was intact, without any connection to the maxillary sinus.

After fixation in 4% buffered formalin, the 2 molars were embedded in methylmethacrylate and cut horizontally into approximately 400-μm-thick ground sections using a slow-speed diamond saw (Varicut VC-50; Leco, Munich, Germany). After mounting onto acrylic glass slabs, the sections were ground and polished to a final thickness of ~100 μm (Knuth-Rotor-3; Struers, Rodovre/Copenhagen, Denmark). The surface of every other ground section was stained with toluidine blue/ McNeal, and the remaining sections were left unstained and observed under polarized light.

**DIAGNOSIS AND MANAGEMENT**

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**Fig. 6.** Photomicrograph illustrating the typical appearance of the hyperplastic cementum (toluidine blue and basic fuchsin stain, original magnification ×125). Note the layered appearance due to the presence of growth/resting lines and the high number of cementocytes. The innermost cementum layer consists of acellular extrinsic fiber cementum (AEFC). On the cementum surface, numerous past resorptive lesions can be seen. These Howship lacunae are filled with an intensely stained repair tissue. The soft tissue adjacent to the cementum layer is heavily infiltrated with inflammatory cells. The termination of a long junctional epithelium (LJE) extending from a pocket epithelium is shown (arrow). D = dentin.

**Fig. 7.** Photomicrograph showing numerous small Howship lacunae in the interradicular area. Note that the resorption cavities are filled with bacterial plaque adjacent to inflammatory cells (toluidine blue and basic fuchsin stain, original magnification ×200).

**Fig. 8.** At some sites, the contour of the root surface was very irregular. This irregularity was partly due to the presence of cementicles which were fused with one another and with the cementum surface. The region illustrated here originates from the interradicular area, where the hyperplastic cementum created an environment inaccessible for root instrumentation to remove the biofilm. The cementum surface is heavily covered with bacterial plaque, and inflammatory cells are seen next to it (toluidine blue and basic fuchsin stain, original magnification ×160).
Reconstruction of the serial ground sections revealed that the apical three-fourths of the surfaces of the 3 roots were covered with a thick, hyperplastic cementum layer (Fig. 5). Whereas the palatal and distobuccal roots were fused together via cementum just under the roof of the furcation, all 3 roots were fused together in the apical root portion. The cementum thickening was observed almost circumferentially in all 3 roots, except at the palatal aspect of the palatal root. The thickness of the hyperplastic cementum ranged from 1 to 2 mm. Characteristic features of the hyperplastic cementum were its layered appearance, caused by numerous growth/resting lines, and its high number of cementocytes (Fig. 6). The innermost cementum layer along the coronal three-fourths of the roots consisted of acellular extrinsic fiber cementum. The bulk of cementum was made up of cellular intrinsic fiber cementum. Crevicular epithelium and a heavy inflammatory cell infiltrate were observed reaching the apical root surfaces. Superficial resorption cavities (Howship lacunae) filled with a strongly stained mineralized repair tissue were regularly observed. Occasionally, Howship lacunae were covered with bacterial plaque, particularly in the furcation area (Fig. 7). Plaque on the root surface was observed all along the tooth surface, from the crown down to the apical root portion (Fig. 8). On the root surface, occasional cemental protuberances seemed to consist of cementicles that were fused with one another and with the cementum surface. The histopathologic analysis of the soft tissue showed signs of chronic inflammation, resembling granulation tissue.

Based on the clinical, radiographic, and histopathologic findings, the final diagnosis was hypercementosis. After this diagnosis, a panoramic radiograph (Fig. 9) was obtained for further screening. No evidence of hypercementosis was detected in any of the other quadrants.

The patient reported minimal postoperative pain and swelling. Wound healing of the defect was uneventful. At the 4-month follow-up visit, soft tissue healing was noted. Remaining exposed hard tissue distal to the root of the second premolar was polished with round burs under water cooling. A thorough plaque control program was established, and the patient was informed about the unclear prognosis of both premolars.

At the 1-year follow-up, progressive gingival recession of the second premolar with exposure of the hypercementosis on the buccal side and a 6-mm exposure of the root surface distally could be seen. The patient's plaque control was good, with no signs of suppuration or advanced gingival inflammation. Vertical tissue deficiency distal to the second premolar was noted. The long-term prognosis of the upper left premolars is guarded, because bacterial contamination of the root surfaces and progressive tissue loss cannot be prevented.

**DISCUSSION**

The literature on the etiology and pathogenesis of hypercementosis is sparse. It is known that root cementum is a dynamic tissue that shows progressive thickening over life, with variations among tooth groups and tooth surfaces. In a study of >22,000 patients with an average age of 42 years, single-tooth hypercementosis was noted radiographically in 1.7% of the subjects. Mandibular molars were more commonly affected than those of the maxilla. The cementum has a tendency to reduce root surface concavities; therefore, thicker cementum layers are found in root surface grooves and in the furcation of multirooted teeth. Interestingly, non-functioning impacted teeth generally have thicker cementum than teeth in occlusion, and the structural architecture is different.
Whether the cementum of human teeth, like bone, has a dynamic response to tensile forces is still a matter of controversy. In case of a localized noninfectious trauma to a root, spontaneous healing by migration of the adjacent cementoblasts and reparative cementum formation occurs. If the trauma is more severe or has an infectious component, root resorption processes can take place, but it has never been shown that hypercementosis is a direct or clear consequence of a localized or severe trauma.

Tooth fusions are defined as a union of originally separated tooth buds at the crown level or at the crown and root level, and are considered to be a developmental disorder of unknown etiology. If union of teeth occurs only at the level of cementum, the term “concrescence” is used, and it is unknown if this is a developmental disorder or a later union. One case report showed hypercementosis of the roots of a second upper molar and an atypical fusion to the crown of an unerupted third molar.

In the literature, cases of generalized hypercementosis are rare. Heredity and systemic disturbances are considered to be etiologic factors for these generalized cases. Thyroid goiter, rheumatic fever, rheumatoid arthritis, acromegaly, calcinosis, and vitamin A deficiency have all been implicated. An investigation of 104 skulls from a 17th to 19th century cemetery on the Newton Plantation, Barbados, showed extensive hypercementosis, as well as a high skeletal lead content. In Paget disease, the cementum formation can also be disorganized, resulting in hypercementosis. In contrast to other causes of hypercementosis, no periodontal ligament space can be seen in Paget disease, because there exists a fusion of the cementum with the surrounding pathologic bone, resulting in ankylosis and root resorption.

In the present case, no etiologic factor could be found for the extensive hypercementosis. A relationship to the trauma 15 years earlier was critically analyzed, but can only be speculated on. No documents related to this trauma were available, and the patient did not remember any intraoral injuries. The patient affirmed that no tooth dislocation occurred and no restorative tooth treatment had to be performed directly afterward.

The patient did not report a history of periodontitis, and no clinical or radiologic signs of periodontitis were present in the other quadrants. Periodontitis as an etiologic factor in the formation of hypercementosis has never been documented in the literature. Nevertheless, the roughness of the root surface because of hypercementosis could have favored the bacterial contamination leading to localized bone loss with marked gingival recession and overall poor prognosis of the affected teeth. There could be a pathogenetic analogy to enamel pearls provoking periodontal pocket formation. The consistent finding of acellular extrinsic fiber cementum as the innermost cementum layer down to the apical root portion supports the fact that the stimulus for accelerated deposition of cellular intrinsic fiber cementum was not present from the beginning of cementum deposition on the root dentin.

The final diagnosis of hypercementosis in this case was challenging because of the atypically large area of involvement, including multiple teeth, as well as secondary inflammation of the marginal and apical periodontal spaces.

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