CASE REPORT

Simultaneous presentation of focal cemento-osseous dysplasia and simple bone cyst of the mandible masquerading as a multilocular radiolucency

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The fibro-osseous lesions represent a large group of disorders that have many common characteristics including clinical, radiographic and microscopic features. Although most are of unknown aetiology, some are believed to be neoplastic and others are related to metabolic imbalances. It is not unusual to see these lesions presenting with a range of radiographic appearances, causing considerable diagnostic confusion owing to their similar histology. This case report presents a histologically benign cemento-osseous lesion that is radiographically consistent with a focal cemento-osseous dysplasia, along with a concomitant simple (traumatic) bone cyst within the right mandibular alveolus, with the initial presentation as a multilocular radiolucency. Pertinent literature is reviewed.


Keywords: simple bone cyst; focal cemento-osseous; dysplasia; biopsy; multilocular radiolucency

Introduction

Bone dysplasias constitute a group of conditions wherein normal bone is replaced with fibrous tissue containing abnormal bone or cementum.1 Benign fibro-osseous lesions are disturbances in bone metabolism where normal bone is replaced by a connective tissue matrix that then gradually develops cemento-osseous tissue.2 Lesions in this category include fibrous dysplasia, focal and florid cemento-osseous dysplasias as well as periapical cemental dysplasia. Cemento-ossifying fibroma, although classified as and behaving like a benign neoplasm, is often grouped with the fibro-osseous lesions due to comparable histology.3 Clinical and characteristic radiographic appearances often suffice for the initial differential diagnosis of these lesions. In the absence of pain, dysfunction or disfigurement, the lesion may be followed up clinically and radiographically to appreciate any changes over time.

Benign fibro-osseous lesions associated with simple bone cysts have been reported previously and may alter or confound the radiographic appearance of these benign fibro-osseous lesions.3,4 The pathogenesis of simple bone cysts is largely unknown. Venous obstruction and blockage of interstitial fluid drainage, in an area of rapidly growing and remodelling cancellous bone, may play an important role in the formation of the simple bone cysts. There is still no definitive evidence available that could explain the formation of these lesions. Benign fibro-osseous lesions that present an atypical radiographic appearance require a detailed clinical, radiographic and laboratory workup to arrive at a diagnosis.4,5 An appropriate treatment plan can be developed following the diagnosis.

Case report

A 41-year-old African-American woman presented to the division of Oral Diagnosis, New Jersey Dental School (NJDS) for a routine dental check up and was referred to the Oral and Maxillofacial Radiology clinic for a full mouth radiographic examination. Her medical history was significant only for hypertension, which, according to the patient, was well controlled with medication. During the initial review of the full mouth radiographic series, a large radiolucent lesion was observed on the edentulous right mandibular premolar-molar region in the periapical views.
The lesion extended mesiodistally from the distal of the root of the second premolar to the mesial of the third molar root and superioinferiorly from the crest of the edentulous alveolar ridge to the inferior alveolar canal. The lesion had distinct borders and a somewhat multilocular appearance. The locules were continuous in some areas, appearing to have coalesced. The bone adjoining the borders of the lesion was sclerosed in several places, giving a pseudo-corticated appearance (Figure 1). An occlusal view (Figure 2) revealed no expansion, thinning or perforation of the buccal and lingual cortices. A panoramic view (Figure 3) was taken to see the entire extent of the lesion. The lesion measured approximately 5 cm × 8 cm on the panoramic radiograph and extended inferiorly beyond the inferior alveolar canal just above the inferior cortex of the mandible.

The radiographic differential diagnosis included odontogenic keratocyst, ameloblastoma, odontogenic myxoma, cemento-ossifying fibroma, central giant cell granuloma and a focal cemento-osseous dysplasia. Clinically, the edentulous ridge on the side of the lesion was non-expansile and had a normal appearance (Figure 4). The non-expansile nature of the lesion decreased the likelihood of the ameloblastoma, cemento-ossifying fibroma and perhaps even giant cell granuloma, since these would probably be expansile given the overall size of the lesion. The patient was referred to oral and maxillofacial surgery for further work-up and treatment. After a positive aspiration yielded a small amount of straw-coloured aspirant (Figure 5), an incisional biopsy of the area was performed which revealed a mixture of gritty bone-like material within an empty cavity. The tissues were submitted for histopathology.

The histological examination of the specimen revealed multiple irregular pieces of hypercellular fibrous connective tissue. Embedded in the tissue were islands of woven bone trabeculae, which were often bordered by thick osteoid rims and prominent osteoblasts. Also present were widely dispersed ovoid calcified structures resembling cementum (Figure 6). One region exhibited remnants of a partially collapsed cavity lined by slightly compressed connective tissue and containing traces of an amorphous proteinaceous substance with admixed erythrocytes (Figure 7). There was no evidence of epithelium. A non-specific diagnosis of benign cemento-osseous lesion was rendered. The diagnosis of simple bone cyst was arrived at based on the surgical findings (empty cavity), radiological findings, the straw coloured aspirant and the lack of epithelial lining histologically.

Based on this diagnosis, a curettage of the entire lesion was performed under local anaesthesia. During the surgery, the lesion was noted to be in close proximity to the inferior alveolar canal, as was expected from the appearance on the radiographs. Care was taken to avoid damage to the inferior alveolar canal. However, the patient developed moderate paraesthesia over the distribution of the inferior alveolar nerve on the right side. On the 3-month follow-up, the patient reported a gradual improvement in sensory function over the right chin and lower lip. Improvement continues to be monitored.
Figure 3  The full extent of the lesion mesiodistally and superior–inferiorly can be seen on the panoramic radiograph of the patient.

Figure 4  Pre-operative clinical appearance of the edentulous mandibular ridge on the right side.

Figure 5  Pre-operative aspirant from the lesion.

Figure 6  Portions of the lesion exhibited a hypercellular dense fibrous connective tissue stroma containing irregular islands of bone (asterisk) and numerous ovoid cementum-like structures (arrow), consistent with cemento-osseous dysplasia (haematoxylin and eosin 100×).

Figure 7  A component of the lesion consisted of a partially collapsed cavity containing erythrocytes admixed with a proteinaceous fluid (asterisk). This cavity lacked an epithelial lining (haematoxylin and eosin 100×).
Discussion

Fibro-osseous lesions of the jaws are of particular interest to oral and maxillofacial surgeons, radiologists and pathologists, as they emphasize the crucial role of these specialists in the diagnostic process and therapy. Since fibro-osseous lesions tend to have a wide range of presenting radiographic appearances, familiarity with the different presentations of these benign lesions is imperative for appropriate treatment planning. For example, periapical cemental dysplasia, focal and florid cemento-osseous dysplasias can generally be identified on routine radiographic examinations because of classic radiographic features and the fact that patients are usually asymptomatic. Therefore, a biopsy is not usually called for. Biopsy is necessary in cases where the differential diagnosis includes lesions with common radiographic and clinical features, but that differ in histology and prognoses. Another example is an early cemento-ossifying fibroma that can easily be confused on clinical and radiographic grounds with focal cemento-osseous dysplasia. Beginning with a radiolucent stage, these cemento-osseous lesions normally form osteoid and cementoid tissues and their appearances change over time. The radiolucent stage is replaced by a mixed opaque-lucent stage, which may yield to a completely opaque appearance.5

A simple bone cyst can be found in association with benign fibro-osseous lesions such as cemento-osseous dysplasia and fibrous dysplasia.1,2 Melrose and co-workers8 were the first to observe this association in their series of 34 cases where 14 patients had concurrent, biopsy-proven simple bone cysts. Aspiration of these cavities will sometimes yield a small amount of straw-coloured or sero-sanguinous fluid,9 as was true in this case (Figure 5). Horner and Forman10 confirmed the association of simple bone cysts occurring with benign fibro-osseous lesions in the jaws when they identified these co-existing lesions in four of their patients who were primarily of West African and West Indian origin.

Plain radiography and/or pantomographic radiography are the examinations of choice in the diagnosis of benign cemento-osseous lesions as well as simple bone cysts. CT and MRI are not required for diagnosis of maxillary and mandibular lesions but can be used for evaluation of anatomically complex areas such as pelvis or spine. Simple bone cysts show little or no uptake of tracer material in radionuclide bone scans unless they have been traumatized.9 While not routinely used or advocated by the authors at this institution, dynamic CT scanning may help in differentiating a fluid containing simple bone cyst, which is avascular, from other solid benign bone lesions that demonstrate varying degrees of vascularity. Similarly, MRI can confirm the presence of fluid within a simple bone cyst. Uncomplicated simple bone cysts have low signal intensity on T1 weighted images and high signal intensity on T2 weighted images. Lesions that have pathological fractures have heterogeneous signal intensities on both T1 and T2 weighted images because of the bleeding within the cyst. When employing MRI with gadolinium diethylene-triamine pentaacetic acid (DTPA) enhancement in other bones of the skeleton, simple bone cysts demonstrate enhancement with focal, thick peripheral, heterogeneous or subcortical patterns. Septations within the lesion may be observed on MRI and may not be visualized on radiographs.

Radiographically, this mandibular lesion had the appearance of a multilocular lucency with scalloped borders. There were incomplete radiopacities resembling septae within the lesion, which was in agreement with prior published work about the nature of these apparent septae.11 The buccal and lingual cortical borders of the mandible appeared unaffected. In the present case, the radiographic appearance of the lesion proved to be atypical and was suggestive of a cystic or neoplastic lesion such as the odontogenic keratocyst, ameloblastoma, or odontogenic myxoma. Corticated borders, expansile, multilocular appearance and the location within the posterior body of the mandible usually characterize these lesions.

Ameloblastoma, odontogenic keratocyst and odonto-genic myxoma were included in the differential diagnosis due to their characteristic multilocular radiolucent appearance with well defined borders, as well as the extent of these lesions within the mandible. Generally these lesions occur in the posterior mandible.

The odontogenic keratocysts tend to have septae within the large lucent lesions. Although commonly appearing in the anterior mandible, the central giant cell granuloma may also be included in the differential diagnosis of multilocular radiolucent lesions. It exhibits a painless expansion of bone and radiographically appears as either unilocular or multilocular lucent defect with well-delineated margins that are non-corticated. Microscopically, the lesions of central giant cell granuloma can be easily differentiated from other lesions on the differential diagnosis by the presence of cellular dense fibrous connective tissue stroma with large numbers of extravasated erythrocytes and varying amount of multinucleated giant cells.12

Cemento-ossifying fibroma is an encapsulated benign neoplasm that appears almost exclusively in the facial bones during the third and fourth decades of life. It tends to have mixed radiolucent densities that are somewhat amorphous in nature.13 Cemento-ossifying fibromas appear radiographically as dense opacities with well-defined margins surrounded by a thin zone of luency and may have a close resemblance histologically to benign fibro-osseous lesions.14–16

When simple bone cysts are found in association with focal or periapical cemento-osseous dysplasia, they can be found anywhere in the tooth-bearing regions. The simple bone cysts that occur without any association with cemento-osseous dysplasia tend to heal much better after surgery than those associated with cemento-osseous dysplasia.2 The histological features of simple bone cysts are mostly non-specific, and when a tissue is submitted from this lesion, a microscopic examination will reveal only a strip of fibrous connective tissue, occasionally with an associated rim of bone.

Radiographic appearances of periapical or focal cemento-osseous dysplasia differ substantially depending on the stage and the site of the lesion as well as other
simultaneously occurring processes. The authors believe that the association between the simple bone cyst and focal cemento-osseous dysplasia is responsible for the unusual radiographic presentation seen in this case. This should be kept in mind when formulating the differential diagnosis of lesions with similar radiographic presentations. Generally, no specific entity can be diagnosed based upon the information obtained solely from the clinical, radiographic or histological examinations. It is essential that the oral and maxillofacial surgeon, the radiologist and the pathologist integrate all relevant and available information to come up with a correct diagnosis and appropriate disease management.

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References