An Interdental Radiolucent Lesion of Mandible: A Case Report and Differential Diagnosis

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Abstract

Aim: The present article discusses a case which was an accidental finding in uncommon location.
Case Report: A 70-year-old apparently healthy woman presented with proximal caries on mesial surface of canine and exhibited mild sensitivity to percussion. An intraoral periapical radiograph demonstrated interdental bone loss along with a diffuse unilocular radiolucent lesion between the canine and premolar region. Pathologies such as Radicular cyst, Lateral Periodontal Cyst, Collateral Keratocystic Odontogenic Tumor (KCOT), Squamous Odontogenic Tumor (SOT), Central Giant Cell Granuloma (CGCG), Extrafollicular Adenomatoid Odontogenic Tumor (AOT), other possible odontogenic and mesenchymal lesions with similar presentation in the anterior jaw were considered and discussed in our differential diagnosis.
Conclusion: After histopathological examination of the incisional biopsy, the lesion was diagnosed of KCOT. The lesion was treated with Carnoy's solution prior to surgical enucleation. The patient had been under regular follow-up for 2 years and showed no recurrence.

KEYWORDS: Interdental radiolucency; Keratocystic Odontogenic tumour; Lateral Periodontal cyst.

Introduction

CASE REPORT

A 70-year-old woman presented to the oral and maxillofacial surgeon with the chief complaint of food impaction and sensitivity in the mandibular left canine and premolar region since 20 days. Intraoral examination revealed proximal caries on mesial surface of canine and exhibited mild sensitivity to percussion. Oral hygiene status was poor. Mild generalized gingival recession and a probing depth of 5 mm was evident distal to canine with no mobility of teeth. Patient was in apparent good health and her medical and social history was noncontributory. She was not taking any prescribed medications.

No remarkable finding seen on extraoral head and neck examination. There was no evident lymphadenopathy. An intraoral periapical radiograph was taken at the dental office which demonstrated interdental bone loss along with a unilocular radiolucent area between the roots of canine and premolar (Fig 1). The lesion was diffuse, extending from the alveolar crest to the apex of the roots. Partial loss of lamina dura was present on distal and mesial aspect of canine and premolar respectively.

DIFFERENTIAL DIAGNOSIS

Based on the clinical and radiographic interpretation, radicular cyst was initially

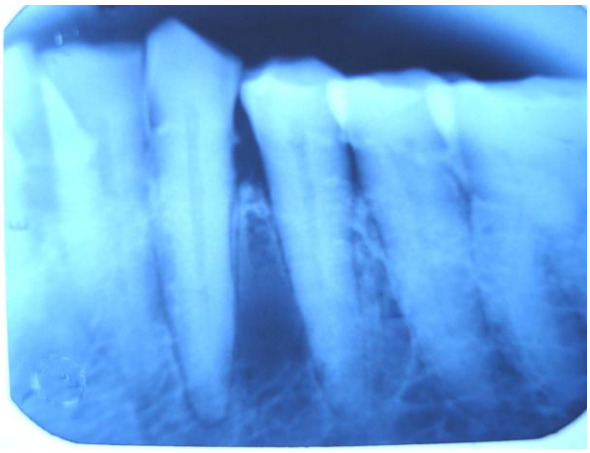

Figure-1 A diffuse unilocular radiolucency present between roots of canine and premolar.

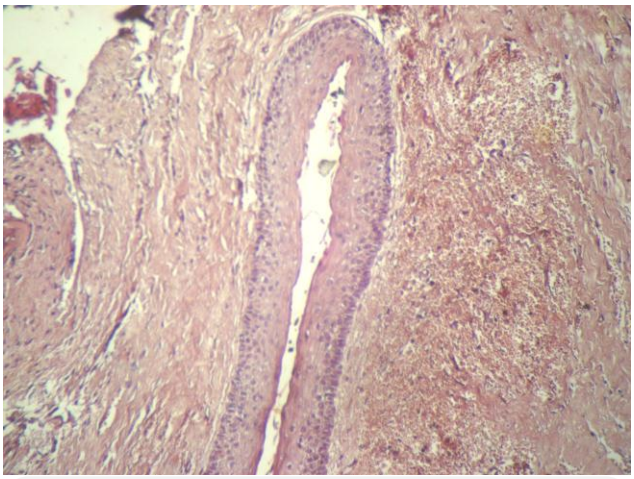

Figure-2 Cystic space lined by parakeratinized stratified squamous epithelium overlying thin connective tissue wall (hematoxylin and eosin, magnification x100).

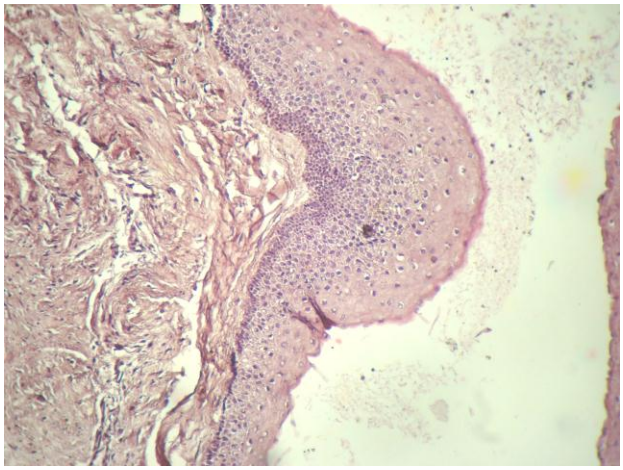

Figure-3 Increased epithelial thickness in the lining (hematoxylin and eosin, magnification x100).

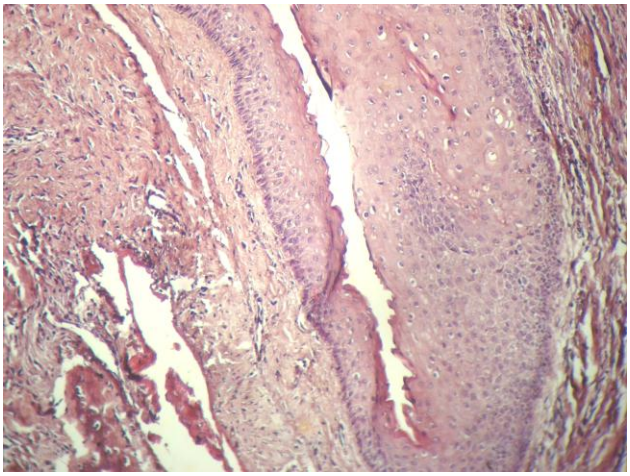

Figure-4 Parakeratotic cells towards the lumen showing corrugation (hematoxylin and eosin, magnification x100).

considered in the differential diagnosis, however, it was not favoured as the associated tooth showed early dentinal caries and positive for vitality test. The most common lesion at this site to be considered in the differential diagnosis was lateral periodontal cyst, which is rare amongst developmental odontogenic cysts. It is more commonly seen in adults, predominantly 6th decade with no gender predilection. Clinically, it presents as an asymptomatic gingival swelling, commonly affecting mandible, localized in canine and premolar region.[1,2] Radiograph shows a round or oval radiolucency often less than 1 cm in diameter usually with sclerotic border. The cyst lies somewhere between the apex and the apical margin of the tooth.[2] Loss of lamina dura and periodontal space may be present.[3]

Collateral Keratocystic Odontogenic Tumor (KCOT) is the next common lesion seen in this location. This tumor shows a bimodal age distribution occurring commonly in the mandible and very rarely in the anterior maxilla. Clinically, it manifests as pain, swelling, expansion of the cortical bone and/or occasionally parasthesia of the lip or teeth. Radiographically, KCOT manifest as unilocular or multilocular radiolucency with smooth or scalloped margins, adjacent to the roots of teeth, usually in mandibular premolar region. The margins may be distinct or diffuse.[2, 4]

Squamous Odontogenic Tumor (SOT) usually occurs intraosseously and probably develops in the periodontal ligament between the roots of vital erupted permanent teeth.[5, 6] Mobility of teeth, local pain, swelling of the gingiva, osseous expansion or mild erythema may be observed.[5] It has been found in almost all age groups and does not seem to have a predilection for any site or gender.[1]But according to Philipsen and Riechart, it is commonly seen in third decade with male predominance[7] and mandible is affected more than

maxilla.[7,8] The characteristic radiographic feature is that of a triangular shaped or a semicircular radiolucency associated with the roots of the erupted teeth.[1,5,7,8]

Central Giant Cell Granuloma (CGCG) is a benign proliferative lesion with no defined etiology. It is seen in a wide age range, commonly in young females below 30 years of age with predilection for mandibular anterior segments. The lesion may present no signs and symptoms, may be discovered accidentally, but sometimes show cortical expansion, mobility, displacement of teeth and root resorption of the associated teeth. It is a unilocular or multlocular radiolucent lesion with smooth or ragged border and usually non- corticated but sometimes it may show faint trabeculae.[4]

Extrafollicular Adenomatoid Odontogenic Tumor (AOT) was also considered as one of the radiographic differential diagnosis. 30% of the central AOTs are extracoronal (extrafollicular) and demonstrate a relationship to the roots of the adjacent or nearby teeth that range from lateral or interproximal to periapical to no relationship at all. Among the extracoronal cases where the exact location was reported, 89% of AOTs occurred adjacent to a permanent cuspid. They present as well demarcated, almost always unilocular radiolucency that generally exhibits smooth corticated border.[9] However, in contrast to present case, this lesion is most commonly seen in anterior maxilla of younger age groups. It is usually asymptomatic but however, gingival swelling or jaw enlargement is seen.[7, 9]

Despite a small unilocular radiolucency few other possible lesions with similar presentation in the anterior jaw such as glandular odontogenic cyst, central odontogenic fibroma, ameloblastoma, central odontogenic myxoma, ameloblastic fibroma and benign central mesenchymal tumors like solitary neurofibroma and neurilemmoma were also considered in our differential diagnosis.

Glandular Odontogenic Cyst (GOC) is an uncommon jaw cyst of odontogenic origin seen in middle age with slight male predilection.[2, 10] Although mandibular anterior region is the most common site of occurrence, few cases from canine to molar region have also been reported.[2] The lesion is asymptomatic, but occasionally accompanied by pain. It is a uni or multilocular lesion with either smooth or scalloped margins.[2, 10] The lesion has shown local aggressiveness and has a recurrence potential.[11]

Central Odontogenic Fibroma (COF) unlike other odontogenic cysts and tumors, has a predilection for anterior mandible in females.[7, 4, 12] Clinically, it is asymptomatic except for swelling of the jaws and typically manifests as a unilocular radiolucency.[4,12]

In Ameloblastoma, although posterior mandible is commonly affected, according to Reichart and Philipsen, 7.7% of this lesion occurs in canine and premolar area. Radiographically, smaller ameloblastomas often present as a unilocular radiolucency with sclerotic borders.[7]

Central odontogenic myxoma may occur anywhere in jaws but have predilection for molar and pre molar region of the mandible and maxilla. Radiographic feature usually range from small unilocular lesion to large multilocular radiolucencies, which often displace teeth or, less frequently, resorb roots. Cortical expansion and perforation is common.[4, 7, 12]

Ameloblastic fibroma is rarely seen in the anterior region of the jaws. Clinically it is a painless, expansile and slow growing lesion. 75% of the cases are associated with impacted teeth. This lesion appears as unilocular or multilocular radiolucency with a smooth well defined outline and often with a sclerotic opaque border.[7, 13]

Benign neural tumors such as neurilemomma and neurofibroma, which are occasionally seen as intra osseous lesions, produce well demarcated or poorly defined unilocular or multilocular radiolucencies. These lesions are usually associated with pain and parasthesia. Solitary neurofibromas are rare and are usually a component of neurofibromatosis.[14]

DIAGNOSIS AND MANAGEMENT

Histopathological examination of the incisional biopsy tissue revealed cystic lumen lined by parakeratinized stratified squamous epithelium overlying a thin, friable connective tissue wall (Fig 2). The epithelial lining was approximately six to eight cell layer thick throughout but focal areas showed increase in cell layer thickness (Fig 3). The basal columnar and cuboidal cells were hyperchromatic and palisaded. The cells towards the lumen were flattened, parakeratotic with a corrugated appearance (Fig 4). The epithelium and connective interface was flat and showed its detachment in few areas. The underlying connective tissue wall was fibrous and almost devoid of inflammatory cells. The final diagnosis was given of odontogenic keratocyst, collateral variety. Gorlin-Goltz syndrome was ruled out. The lesion was treated with Carnoy's solution prior to surgical enucleation. The patient had been under regular follow-up for 2 years and showed no recurrence.

DISCUSSION

Odontogenic keratocyst (OKC) is classified as a developmental cyst and comprises approximately 11% of all the cysts of the jaws.[15] It is one of the most aggressive odontogenic cysts of oral cavity.[16] Philipsen introduced the term in 1956. In 1967, Toller suggested that the OKC may best be regarded as a benign neoplasm rather than a conventional cyst based on its clinical behavior. Recently, a wealth of clinical and molecular evidence has

indicated that odontogenic keratocyst (OKC) should now be regarded as benign cystic neoplasm. In the years since, published reports have influenced WHO to reclassify the lesion as a tumour. Several factors form the basis of this decision. Behaviourwise, the KCOT is locally destructive and highly recurrent. Histopathological studies, such as that by Ahlfors and others show the basal layer of the KCOT budding into connective tissue. In addition, WHO notes that mitotic figures are frequently found in the suprabasal layers. Genetically, PTCH ("patched"), a tumour suppressor gene is involved in both NBCCS and sporadic KCOTs, which occurs on chromosome 9q22.3-q31. Normally, PTCH forms a receptor complex with the oncogene SMO ("smoothened") for the SHH ("sonic hedgehog")ligand. PTCH binding to SMO inhibits growth-signal transduction. SHH binding to PTCH releases
this inhibition. If normal functioning of PTCH is lost, the proliferation-stimulating effects of SMO are permitted to predominate.[17]The majority of patients are in the age ranges of 20-29 and 40-59 but ranges from 5 to 80 years have been reported.[16] The distribution between the sexes varies from equality, to a male to female ratio of 1.6:1, except in children. Lesions found in the children are often reflective of multiple odontogenic keratocysts as a component of the nevoid basal cell carcinoma syndrome. There are no characteristic clinical manifestations of the keratocyst, among the most common features are pain, soft- tissue swelling and expansion of bone, drainage and various neurological manifestations such as parasthesia of the lip or teeth.[4] The OKC tends to expand in the medullary cavity and clinically observable expansion of the bone occurs late. It may occur in any part of the upper and lower jaw, although there is considerable predilection for mandibular 3rd molar region and ramus, they may also occur in maxilla.[2] There has been a great deal of interest in OKC since they are known for their varied origin, debated development, peculiar behaviour, unique tendency to recur following surgical treatment. Typical radiographic features such as scalloped margins and multilobular or multilocular / unilocular radiolucency are indicative but are not unequivocal.[18] Main has referred to a variety of OKC radiographically that include envelopmental, replacement and extraneous. Term collateral OKC is proposed for OKCs adjacent to roots of the teeth usually in mandibular premolar region, which are indistinguishable radiologically from lateral periodontal cyst. In Forsells series the collateral type occurred in 19% of OKCs.[2] The present case report highlights one such rare radiographic variant of OKC, which was an accidental finding with no clinical presentation. Treatment of OKCs remains a controversial subject. A review of literature suggests that recurrence rate is relatively low with aggressive treatment, whereas more conservative methods tend to result in more recurrences. Enucleation is a commonly used method for surgical treatment of OKCs. The primary advantages of enucleation are the complete removal of the cyst and a thorough histopathologic examination of the lesion. However, surgical complications include compromised vitality of adjacent teeth, nerve damage, and encroachment on anatomic structures, such as the maxillary sinus, floor of the nasal cavity, and the mandibular canal. Recurrence has been suggested to be a consequence of technical difficulties in complete removal because of the thin cystic epithelium or because of the location. Excision of the oral mucosa overlying areas of cortical perforation should be performed. Treatment of the cyst with Carnoy's solution for 10-15 minutes before enucleation is helpful to prevent any remnants that are left from developing into recurrence, but it may also damage adjacent bone and nerve fibers. If the inferior alveolar nerve is visible in the bony cavity after enucleation, Carnoy's solution should not be directly applied to this area or applied beyond 3 minutes. Resection should be considered for treatment of a recurrent OKC and, when performed, should extend beyond the greatest extent of the lesion to ensure complete removal of remaining satellite cysts or epithelial remnants of cyst wall.[15] Resection of the jaw results in the lowest recurrence rate. However, considering the radical nature of the procedure, unless resection is necessary, it is acceptable to use enucleation in combination with Carnoy's solution and peripheral ostectomy or marsupialization with cystectomy.[17]
As research continues, treatment may become molecular in nature. Zhang and others postulate that antagonists of SHH signalling factors could effectively treat KCOTs. Their suggested strategies include the reintroduction of a wild-type form of PTCH, inhibiting the SMO molecule by synthetic antagonists and suppressing the downstream transcription factors of the SHH pathway. They suggest that intracystic injection of an SMO protein-antagonist has the greatest potential as a future treatment option.[17]

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