Multiple Multilocular Dentigerous Cysts with Intra-osseous and Extra-osseous Third Molar Displacement: A Case Report

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ABSTRACT

The dentigerous cyst with extra-osseous tooth displacement is a rare presentation for one of the most common odontogenic (developmental) cysts. Association with an impacted third molar is the most common finding for a dentigerous cyst but actual displacement of a tooth into the soft tissue has never been reported. The diagnosis of a dentigerous cyst is usually made as an incidental finding following radiographic investigation. This entity is quite frequently asymptomatic unless secondarily infected. This lesion can cause bone and tooth destruction. The authors discuss the unusual clinical and radiographic presentation, the surgical management and three-year follow-up of a 77-year-old male patient with multiple multilocular dentigerous cysts demonstrating either intra-osseous and/or extra-osseous third molar displacement. In addition, this paper will review histopathologic features and radiographic differential diagnosis of this presenting lesion.

Impacted third molars can be associated with many pathologic entities which include dentigerous cysts, chronic nonspecific inflammatory tissue, odontogenic keratocysts and ameloblastomas. The dentigerous cyst is the most common pathologic lesion associated with impacted third molar teeth.[1] Dentigerous cysts occur at a frequency of 1 to 2 cysts for every 100 impacted teeth.[2] The dentigerous cyst is a developmental cyst of odontogenic origin. This cyst is an epithelial-lined pathologic cavity that encompasses the crown of an impacted tooth. The lining encases the crown at the cemento-enamel

junction and arises from the separation of the follicle from the crown of an unerupted tooth. The cyst lining originates from the reduced enamel epithelium. The exact stimulus for this developmental cyst is unknown.

Dentigerous cysts are frequently discovered when radiographs are taken to investigate failure of tooth eruption, a missing tooth or cause for misalignment. Generally, few symptoms are associated with a dentigerous cyst unless secondarily infected or associated with inflammation (pericoronitis) but on occasion, paraesthesia has been noted.[3] The radiographic appearance of a dentigerous cyst is usually that of an unilocular, non-expansile radiolucency characterized by well-defined borders and is always associated with the crown of an impacted tooth, most often a third molar. Dentigerous cysts range in size from 5mm to 7cm or even larger.

The presentation of multilocular dentigerous cysts is unusual. On rare occasions, multilocular and bilateral dentigerous cysts can occur in syndromic patients, including those with cleidocranial dysplasia and Maroteaux-Lamy syndrome.[4] When evaluating multilocular lesions of the mandible, a differential diagnosis should include multiple odontogenic keratocyst nevoid basal cell carcinoma syndrome (Gorlin-Goltz syndrome) and ameloblastomas.[4] The occurrence of bilateral dentigerous cysts associated with non-syndromic patients is rare. This paper reports an undescribed and extremely unusual finding of multiple multilocular dentigerous cysts with both intra-osseous and extra-osseous third molar displacement in a non-syndromic patient.

CASE REPORT

A 77-year-old white male presented to his dentist in March 1999 for routine dental care. The patient presented with no specific dental complaints. His medical history was non-contributory and no apparent systemic problems were presented except a history of hypertension and generalized arthritis. He was taking no medications.

FIGURE 1—Preoperative panoramic radiograph (March, 1999).

FIGURE 2—Preoperative anterior-posterior cephalometric radiograph (March, 1999).

The right and left mandibular molars and right second mandibular bicuspid tested non-vital.

Clinical evaluation

Clinical examination of the facial region without palpation revealed no obvious asymmetry or facial swelling. Examination of the left submandibular region, with palpation, revealed a 2cm mobile mass. No palpable lymph nodes were noted. Intra-oral examination demonstrated an expansile lesion involving the left and right posterior mandible. There was no trismus, paraesthesia or facial paralysis associated with the expansile regions of the posterior mandible. The dentition was extensively restored with mild to moderate periodontal disease. The patient's oral hygiene was fair. The right and left mandibular molars and right second mandibular bicuspid tested non-vital.

Radiographic evaluation

Radiographic examination of the panoramic film taken on initial presentation (March, 1999) demonstrated heavily restored dentition, missing left maxillary first bicuspid and three areas of significant osseous and/or dental changes (Fig. 1). The left posterior maxilla demonstrated a partially demarcated radiolucent lesion with well-defined posterior borders in association with an impacted third molar. It was associated with the cemento-enamel

junction of tooth #28 and measured 1.8 x 1.0cm. There was a well-defined multiolocular radiolucency of the right mandible associated with a horizontally impacted and posteriorly displaced third molar. The inferior border of the right mandible appeared intact but thin and the lesion measured approximately 7.0 x 3.0cm in greatest dimension. The root of tooth #47 and mesial root of tooth #46 were resorbed and the apex of tooth #45 appeared blunted.

Additionally, a well-defined multilocular radiolucency was noted in the left posterior mandible in association with an impacted third molar that had been displaced inferiorly into the soft tissue of the submandibular region. The buccal cortex of the left mandible demonstrated expansion and the inferior cortex was discontinuous. The radiolucency measured approximately 7.0 x 3.5cm in greatest dimension. There appeared to be some superior expansion and perforation of the superior alveolar ridge distal to tooth #37 (Fig 2). The second molar also appeared to be superiorly displaced and demonstrated apical root resorption. A CT Scan demonstrated a multi-locular expansile lesion involving the left and right posterior mandible with osseous perforations and displacement of teeth (Fig. 3).

The differential diagnosis based on the clinical and radiographic evaluation included dentigerous cyst, odontogenic keratocyst and ameloblastoma. The nature of lesion, risk and benefits of surgery and possible complications, including paraesthesia and fracture were explained to the patient. The patient elected to proceed with the surgical intervention.

Surgical treatment

This 77-year-old otherwise healthy patient was treated under general anaesthesia in a private out-patient non-hospital surgical facility. The impacted left maxillary third molar was surgically removed and the associated lesion was enucleated. The adjacent erupted left maxillary second molar was extracted due to significant bone loss on distal secondary to expansion of the lesion. The impacted right mandibular third molar was surgically removed. The large expansile lesion was enucleated and peripheral ostectomy was performed. The inferior alveolar neurovascular bundle was visualized in the defect and remained intact and uninjured. The non-vital

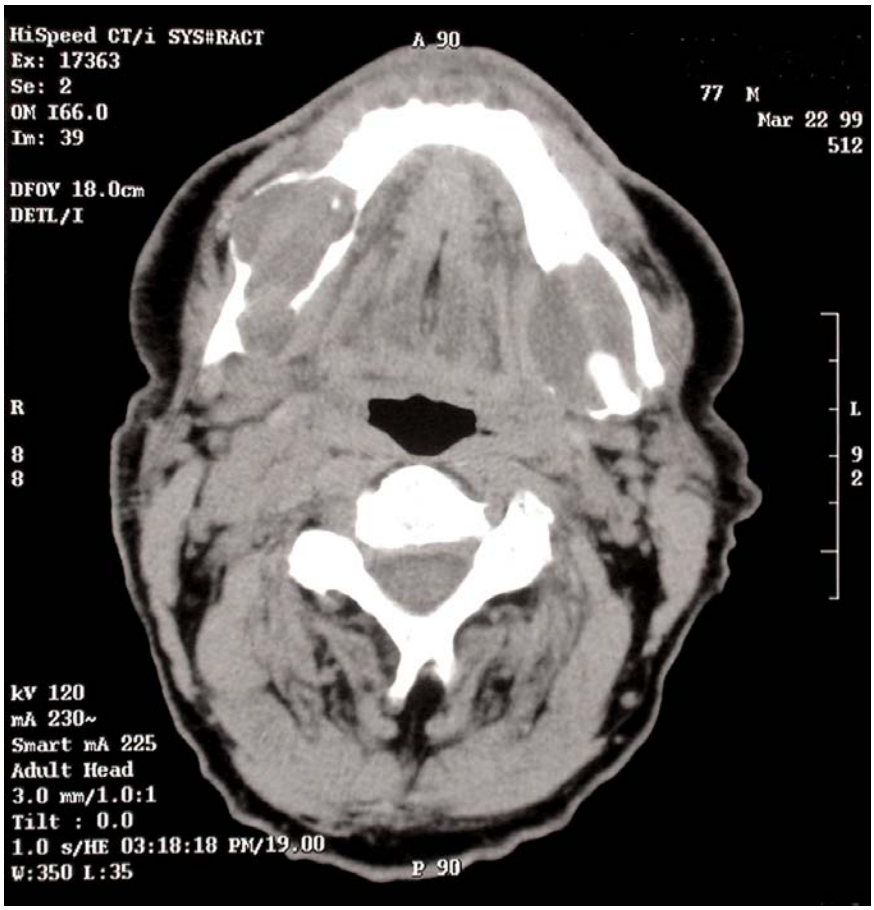

FIGURE 3—Preoperative axial CT scan (March, 1999).

FIGURE 4—Immediate post-operative panaromic radiograph (April, 1999).

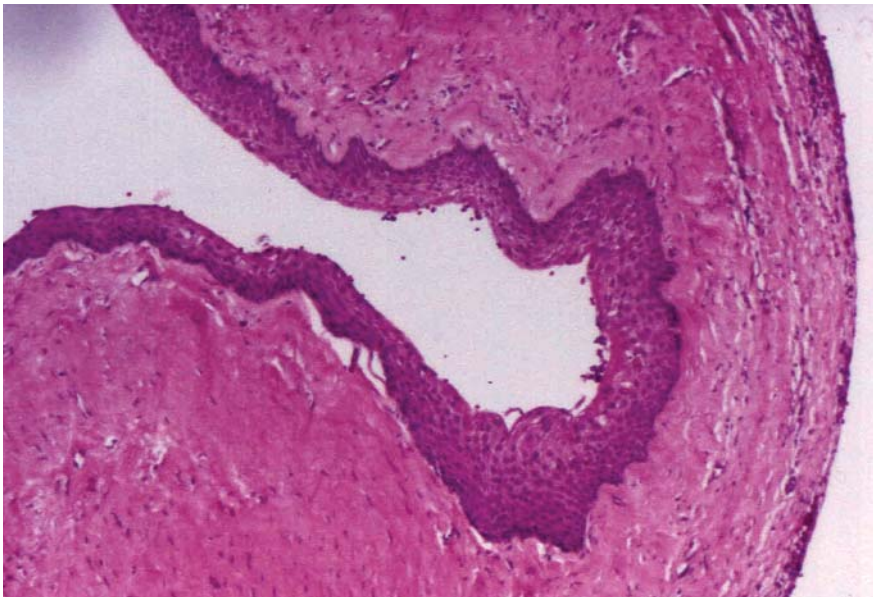

FIGURE 5—Photomicrograph of the wall of a dentigerous cyst exhibiting nonkeratinized stratified squamous epithelium with supporting follicular fibrous connective tissue. This is representative of tissue associated with impacted tooth # 28 (hematoxylin-eosin, original magnification x 63).

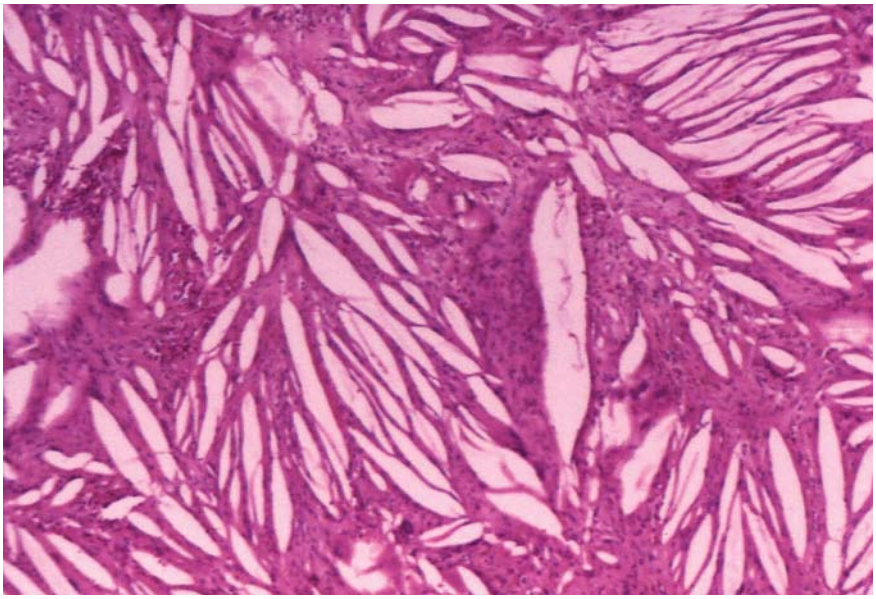

FIGURE 6—Photomicrograph of cholesterol clefts surrounded by multinucleated giant cells and granulation tissue. This is representative tissue from the specimen associated with impacted tooth #28 (hematoxylin-eosin, original magnification x 63).

right mandibular molars and second bicuspid were extracted. A primary closure was established. The left maxillary and right mandibular specimens were sent for histological evaluation.

The displaced extra-osseous mandibular third molar was digitally manipulated back into the large pathologic space through the inferior border defect and extracted intra-orally. The non-vital left first and second mandibular molars were extracted. It was elected at the time of surgery to perform a formal marsupilization of the left mandibular lesion. The inferior alveolar neuro-vascular bundle was not visualized in the defect. Iodophore gauze was utilized to pack open the defect and was removed gradually over an eight-week period. A representative specimen of the left mandibular lesion was sent for histological evaluation.

The patient's intra-operative course and post-operative healing was uneventful. No paraesthesia or mandibular fracture occurred. The patient maintained a blenderized diet for eight weeks and presented for regular progressive dressing removal and routine follow-up. The immediate post-operative panoramic film taken on April 21, 1999, showed the above-mentioned radiolucent areas with a dressing within the surgical site of the left posterior mandible (Fig. 4). A panoramic film taken five months post-operatively showed radiolucent surgical sites of the left posterior maxilla and the right and left posterior mandible (Fig. 7). A panoramic film taken one and a half years post surgically demonstrated healed surgical sites of the left posterior maxilla and right and left posterior mandible. Note the complete reconstruction of the inferior border of the left mandible (Fig. 8). Subsequent radiographic three year follow up demonstrates complete restoration of the osseous contour of the mandible (Fig. 9). The patient currently functions with a mandibular removable prosthesis but desires to proceed with an implant supported prostheses.

Histopathology

Histological evaluation of the specimens was completed. One piece of brown tissue measuring 1.8 x 1.0 x 0.2 cm was submitted from the left posterior maxillary surgical site.

FIGURE 7—Postoperative panorex radiograph (August, 1999).

FIGURE 8—Eighteen months post-operative panaromic radiograph (October, 2000).

Histologic examination revealed non-keratinized stratified squamous epithelium supported by a wall of follicular fibrous connective tissue. Aggregates of cholesterol clefts with surrounding multinucleated giant cells were present. The microscopic diagnosis was consistent with a dentigerous cyst and cholesterol granuloma.

Three pieces of brown tissue were submitted from the left posterior mandibular surgical site. The largest piece was 1.0 x 0.8 x 0.2cm in dimension. Histologically there was a small amount of non-keratinized stratified epithelium with chronically inflamed follicular fibrous connective tissue and cholesterol clefts and necrotic material. The microscopic diagnosis was dentigerous cyst with cholesterol granuloma.

Two pieces of grey tissue were submitted from the right posterior mandible, the largest measuring 5.0 x 1.0 x 0.3 cm. The histologic features included variably thickened non-keratinized stratified squamous epithelium supported by a wall of chronically inflamed follicular connective tissue. There was no evidence of cholesterol granuloma. The microscopic diagnosis was dentigerous cyst.

The two most common neoplasms that can occur in association with dentigerous cysts are ameloblastomas and squamous cell carcinoma.

Discussion

The dentigerous cyst is the second most common benign odontogenic cyst within the oral cavity. Only the radicular cyst occurs with more frequency.[5] Dentigerous cysts are frequently diagnosed in the second and third decades of life. The dentigerous cyst epithelial lining can give rise to neoplasia in cases with long standing chronic inflammation. The two most common neoplasms that can occur in association with dentigerous cysts are ameloblastomas and squamous cell carcinoma.[6] The mean age for identification of malignant disease is 64 years.[7] Therefore, in elderly patients, with large cystic lesions, there is always the concern of malignant transformation which must be ruled out with histological evaluation.

Dentigerous cysts are developmental in origin and commonly associated with impacted third molars that can be displaced in an intra-osseous manner. Radiographically, the dentigerious cyst appears most frequently as a well circumscribed unilocular expansile radiolucency that may or may not exhibit cortication.[8] The posterior area of the mandible is the most common site of dentigerous cysts and the third molar is the tooth that is involved most frequently. Expansile mulilocular dentigerous cysts with bilateral mandibular involvement and tooth displacement have been described in the past.[9] These large expansile lesions can lead to pathologic fracture when significant bone destruction has occurred.

Intra-osseous displacement has occurred where an impacted third molar in the subcondylar region was associated with a dentigerous cyst.[10,11] Furthermore, the presence of unilateral

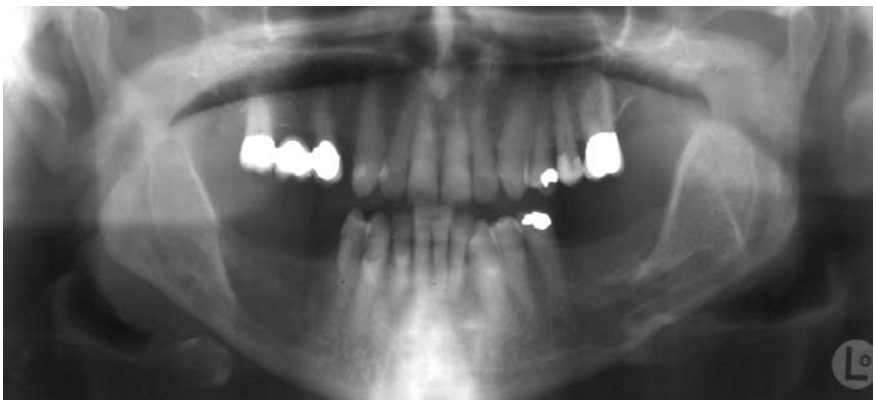

FIGURE 9—Three years post-operative panaromic radiograph (April, 2002).

or bilateral ectopic, impacted mandibular third molars in different regions of the body and ramus has been reported[12-14] but there are no reports in the literature of actual extra-osseous displacement into soft tissue.

The displacement is secondary to slow, progressive cystic enlargement by unicentric expansion because of the hydrostatic pressure of its contents.[15] Two possible theories have been presented on the pathogenesis of the dentigerous cyst. One is that the cyst arises from the collection of inflammatory exudates, which is derived from the obstructed follicular veins of an unerupted tooth and accumulates between the reduced enamel epithelium and the crown of the tooth.[16] An alternative explanation has been that the cyst may originate initially by the proliferation and cystic transformation of islands of epithelium in the connective tissue wall of the dental follicle and this transformed epithelium then unites with the lining follicular epithelium, forming a solitary cystic cavity around the tooth crown.[17]

Different surgical treatment alternatives are available, such as marsupialization, but enucleation of the cyst is the most widely accepted method of treatment. Marsupialization was performed on the left mandible due to the significant size, destruction of the lesion and there was a significant concern for the potential of a pathologic fracture. Bone grafting was never considered as it is well documented by numerous authors that spontaneous bone regeneration is predictable. Spontaneous bone regeneration can occur in large mandibular cysts without the aid of any grafting materials.[18] Extra-oral incision, such as the submandibular approach described in the literature by Kocer, et al.[19] was not necessary as we were able to digitally manipulate the tooth back into the osseous defect. There was no difficulty visualizing the tooth in the defect and completing the removal.

SUMMARY

This report presents the clinical and histological features and surgical treatment of multiple multi-locular radiolucencies involving the mandible. Pathologic changes associated with long standing impacted third molars include displacement and/or destruction of adjacent dental and osseous structures resulting in the potential for pathologic fractures and even malignant change. In this particular case the potential for a bilateral pathologic fracture was significant. This case clearly demonstrates the significant osseous and tooth destruction that can occur with impacted third molars, including third molar intra-osseous and extra-osseous displacement. **OH**

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Oral Health welcomes this original article.

REFERENCES

1. Rakprasitkul S. Pathologic changes in the pericoronal tissues of unerupted third molars. Quintessence Int. 2001 Sep;32(8):633-8.
2. Daley TD, Pringle GA. Relative incidence of odontogenic tumors and oral and jaw cysts in Canadian population. Oral Sur Oral Med Oral Patho 1994; 77:276-80
3. Aziz SR, Pulse C, Dourmas MA, Roser SM. Inferior alveolar nerve paresthesia associated with a mandibular dentigerous cyst. J Oral Maxillofac Surg. 2002 Apr;60(4):457-94.
4. Ko KS, Dover DG, Jordan RCK. Bilateral Dentigerous Cysts-Report of an Unusual Case and Review of the literature. J Can Dent Assoc. 1999 Jan;65(1):49-51.
5. Manganaro AM . The likelihood of finding cult histopathology in routine third molar extractions. Gen Dent. 1998 Mar-Apr; 46(2):200-2.
6. Tsukamoto G, Sasaki A, Akiyama T, Ishikawa T, Kishimoto K, Nishiyama A, Matsumura T. A radiologic analysis of dentigerous cysts and odontogenic keratocysts associated with a mandibular third molar. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2001 Jun;91(6):743
7. Gardner A. A survey of Odontogenic cysts and their relationship to squamous cell carcinoma. J Canad Dent Assoc 1975:49:161-166.
8. Johnson L.M, Saap, J.P., Mcintyre D.N. Squamous Cell Cacinoma Arising in Dentigerous Cysts.
9. Eliasson S, Heimdahl A, Nordenram A. Pathological changes related to long-term impaction of third molars. A radiographic study. Int J Oral Maxillofac Surg. 1989 Aug;18(4):210-2.
10. Ko KS, Dover DG, Jordan RC. Bilateral dentigerous cysts—report of an unusual case and review of the literature. J Can Dent Assoc. 1999 Jan;65(1):49-51. Review.
11. Tumer C, Eset AE, Atabek A. Ectopic impacted mandibular third molar in the subcondylar region associated with a dentigerous cyst: a case report. Quintessence Int. 2002 Mar;33(3):231-3.
12. Bux P, Lisco V. Ectopic third molar associated with a dentigerous cyst in the subcondylar region: report of case. J Oral Maxillofac Surg. 1994 Jun;52(6):630-2. No abstract available.
13. Girod SC, Gerlach KL, Krueger G. Cysts associated with long-standing impacted third molars. Int J Oral Maxillofac Surg. 1993 Apr;22(2):110-2.
14. Callaghan JH. Bilateral impaction of lower third molars in association with dentigerous cyst formation. A case report. Glasg Dent J. 1973 Spring;4(1):36-8.
15. Pressman P, Allen GW. Maxillary sinus mass: the case for the unerupted third molar. Otolaryngol Head Neck Surg. 2000 May;122(5):776.
16. Georgiade NG, Georgiade GS, Harter TB. Solid and cystic tumors of the jaw. Textbook of Plastic, Maxillofacial and Reconstructive Surgery. Baltimore: Williams & Wilkins, 1996:489-506.
17. Shafer WG, Hine MK, Levy BM. A textbook of Oral Pathology. 4th Edition Philadelphia: W.B. Saunders Company, 1983:260-265.
18. Chipasco M. J. Oral Maxillofac Surg Spontaneous Bone Regeneration After Enucleation of Large Mandibular Cysts 58:942-948, ,2000.
19. Kocer Giant Dentigerous Cyst Occupying the Riht Hemimandible Can J Plast Surg Vol 10 July/Aug 2002.

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