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Infrequent radiological features of a dentigerous cyst – A case report

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Keywords

Dentigerous cyst, developmental cyst, enucleation, eosinophilic granuloma, marsupialization

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Abstract

Background: Dentigerous cysts, after the radicular cysts, are the second most common developmental cysts and arise in general between the 2nd and 3rd decade of life, surrounding the neck of an unerupted permanent tooth at its cemento-enamel junction level. Dentigerous cysts are benign cysts and initially asymptomatic, discovered on routine radiographic examination as unilocular pericoronal, well-defined (unless if infected) radiolucency associated with the neck of the crown of an impacted or unerupted tooth. However, if left untreated, they may invade and expand leading to many complications such as facial asymmetry, teeth movement, nerve alterations (rarely) by compression, and pain if secondary infection occurs.

Aim: The aim of this paper was to present a clinical case of a dentigerous cyst in a 39-year-old patient that was treated by complete enucleation of the radiolucent lesion and extraction of associated teeth.

Conclusion: Dentigerous cysts are frequently found in our practice, so it is crucial to follow-up with research to avoid complications and decide on the best treatment according to the individual case of each patient. The original features of this case among the others are the direction of extension of the cyst (mesiodistal extension instead of the vertical extension) and its radiological features which were typically identical to the eosinophilic granuloma.

Clinical significance: This case report shows us the importance of making an appropriate diagnosis through a well done clinical and radiological examination, which will lead us to the appropriate management and treatment.

Introduction

A dentigerous cyst, described first by Paget in 1863, is the most common type of developmental epithelial odontogenic cysts and the second most frequently encountered cystic jaw lesion after the radicular cyst. It encloses the crown of an unerupted tooth (most commonly permanent tooth), at its cemento-enamel junction (CEJ) level by expansion of its follicle.^[1]

In the beginning, differentiation was done depending on the location of the tooth with respect to the cyst between follicular cysts where the whole tooth is encompassed by the cyst and the dentigerous cyst where only the crown is encompassed by the cyst at the CEJ level and the roots are outside. Actually, we call them all dentigerous cysts, despite tooth's inclusion.

Regarding its pathogenesis, it has been suggested that the pressure exerted by an erupting tooth on the follicle may obstruct venous flow inducing accumulation of exudate between the reduced enamel epithelium and the tooth crown.^[2]

While anyone can develop a dentigerous cyst, the literature review showed that they are more common in individuals between 20 and 40 years.^[3]

Although dentigerous cysts may occur in association with any unerupted teeth (occasionally with supernumerary teeth and odontomas), they most often involve more the mandible than the maxilla, the posterior part more than the anterior one, and especially the third mandibular molar. Other relatively frequent sites involved are, in descending order, the maxillary

canine, maxillary third molars, and mandibular second premolars.^[4]

Clinical manifestations

Dentigerous cysts are benign cysts and initially asymptomatic, discovered on routine radiographic examination as unilocular pericoronal, well- defined (unless if infected) radiolucency associated with the neck of the crown of an impacted or unerupted tooth.^[4] However, if left untreated, they may invade and expand leading to many complications such as facial asymmetry, teeth movement, nerve alterations (rarely) by compression, and pain if secondary infection occurs.^[3]

Radiological manifestations and differential diagnosis of dentigerous cysts

Radiologically, an enlarged dentigerous cyst will push the tooth away from its direction of eruption, to the ramus or inferior border (mandibular molars) or to the sinus or nasal floor (maxillary molars and cuspids).^[4]

In a study done by Hyeok *et al.*, they found that downward backward and upward directions are the most common directions of tooth displacement rather than the mesiodistal displacement found in few cases. Cortical bone expansion, loss of bony continuity, and root resorption are less common radiological features of enlarged dentigerous cysts. These features are more found in ameloblastomas and odontogenic keratocysts.^[5]

The radiological differential diagnosis of dentigerous cysts is made between ameloblastomas (unicystic, ameloblastic fibroma, and ameloblastic fibro-odontoma), adenomatoid odontogenic tumors (most of the cases associated with the crown of an unerupted canine), odontogenic keratocysts, and early stages of Gorlin cysts.^[6]

We should also pay attention and differentiate between a large dental follicle and a small dentigerous cyst (specially the canines that may present a large dental follicle). In fact, the dental follicle, a part of tooth germ, originating from odontogenic ectomesenchyme, is physiologically involved in the formation of the future cementum, alveolar bone, and periodontal ligament. The radiographic distinction between an enlarged dental follicle and a small dentigerous cyst can be difficult and fairly arbitrary. However, in general, any radiolucency >3 mm is considered suggestive of cyst formation.

Histological features of dentigerous cysts

Histologically, a normal finding in the stroma of dental follicle is the presence of foci of calcifications. These foci are not seen in dentigerous cysts. In addition to this, the dental sac may show various histopathological changes during tooth development^[7] and a study done by Saravana said that if an odontogenic epithelial lining of a dental follicle changes from reduced enamel epithelium to stratified epithelium, it is indicative of dentigerous like changes.^[8] That is why, when in doubt, a biopsy is imperative.

Malignant transformation of dentigerous cysts

Many articles cited the malignant transformation of dentigerous cysts into ameloblastoma, squamous cell carcinoma, and mucoepidermoid carcinoma if left untreated.^[9] However, this transformation is extremely rare.^[10] The rate of malignant transformation is estimated to be between 0.13 and 3%.^[11]

The aim of this case report is to describe the case of a 39-year-old man who presented to the dental clinic with unfrequent features of a dentigerous cyst as well as to discuss the importance of the appropriate diagnosis which will lead us to the appropriate management and treatment.

Case Report

In November 2018, a 39-year-old man presented to the dental clinic, Zalka, Lebanon.

Chief complaint: Posterior lower right back teeth mobility (Class 2 mobility: >1 mm horizontally, using Miller's classification) that appeared few months before his visit and without any other symptom. The medical history revealed a healthy patient, without any medical or family history. Dental history revealed a nasopalatine cyst surgery removal in July 2018. The overlying mucosa in the region had a normal appearance, but the patient had an overall bad oral hygiene (generalized plaque and bad breath). The teeth 46–47 were mobile with a type 2 mobility [Figure 1].

On palpation, the region was completely asymptomatic and no adjacent lymph nodes were found. Periapical radiographs were taken at this region and showed severe bone loss starting from the distal root of tooth 46 and an extending radiolucent lesion distal to 47. A panoramic and cone-beam computed tomography (CBCT) radiographs were done to localize the lesion extension [Figure 2].

After combining the clinical and paraclinical results, a differential diagnosis was set between:

- Ameloblastoma
- Eosinophilic granuloma (floating teeth – severe lytic bone lesions)
- Dentigerous cyst (less suspicious).



Figure 1: Intra oral view showing normal mucosa appearance

Under local anesthesia, the extraction of teeth 46–47 followed by complete enucleation of the radiolucent lesion was done, copious irrigation with *physiological saline serum* was realized, the flap was adjusted and hermetically closed by means of single sutures (Vicryl® 4/0) and teeth sent to histopathological study [Figure 3]. Two weeks later, we received the results showing on microscopic examination a non-keratinous stratified squamous epithelium. The wall is densified by a plasmocytarich polymorphic leukocyte exudate and the diagnosis was an inflamed dentigerous cyst. On 6 months follow-up, a new CBCT and panoramic were done [Figure 4] which showed early signs of bone healing in the extraction sites.

Discussion

Dentigerous cysts, after the radicular cysts, are the second most common developmental cysts and arise in general between the 2nd and 3rd decade of life^[6] and defined as developmental odontogenic cyst associated with the crown of an impacted

tooth mostly a permanent tooth at its CEJ level (rarely deciduous).^[12]

Although dentigerous cysts are classified as developmental cysts, some investigators cited also the inflammatory dentigerous cysts (IDC) described first in 1980 by Shaw *et al.*

IDC is found in mixed dentition (between 6 and 12 years), more frequently in the mandible than the maxilla, associated with non-vital deciduous teeth roots (mainly lower second deciduous molar) and crown of an unerupted permanent tooth and therefore contributes to its occurrence in mixed dentition mainly^[6] (which was not the case of our 39-year-old patient). Among the symptoms of an IDC (intraoral swelling, mild sensitivity, tooth mobility displacement of adjacent teeth, and bone springiness),^[6] only tooth mobility was observed.

Eosinophilic granuloma was suspected because it is characterized by “floating teeth” due to severe bone loss which was the case on the panoramic view. However, it was ruled out after receiving the histopathological results.

In general, in the posterior mandible, the dentigerous cyst expands superiorly from the third molar site to the ramus or inferiorly along the body of the mandible, without any chief complaint from the patient unless it is infected.^[13] In our case, a mesiodistal extension instead of the vertical extension was noted

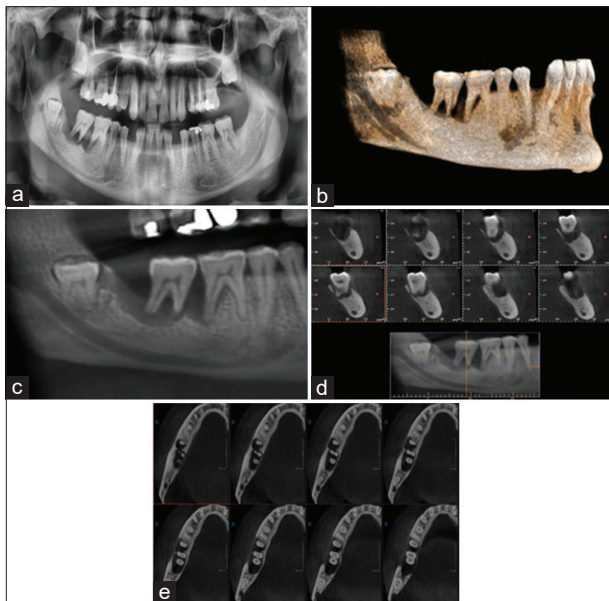


Figure 2: (a) Panoramic X ray demonstrating severe bone loss and radiolucent lesion extending from the distal root of tooth 46 to the wisdom tooth 48 (b) 3D reconstruction (c) Cropped panoramic reconstruction, lower right posterior region. Well-defined low-density periapical image, extending from the distal root of 46 to the ramus. Apical resorption of 47. The teeth seem floating in air. Concerning the tooth 48: According to Pell and Gregory classification: - Depth of insertion: 48 is in a level 3 impaction (fully embedded) -Ramus relationship: class 3 (totally below the angle of the mandible) and finally, for the direction of angulation, we used the winter's classification, in our case, tooth 48 had a vertical angulation. There seems to be a continuity of the low density image mesial to 48 (d) Cross sectional cuts: extension of the lesion - interruption of buccal cortical bone at the level of 47 (e): Series of axial cuts showing the extension of the lesion, interruption of buccal cortical bone and relationship with 48.

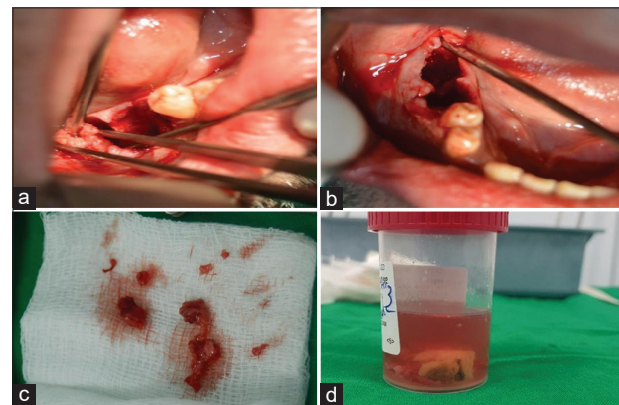


Figure 3: (a) During the lesion removal (b) Complete enucleation. (c) Excised lesion in 3 parts (d) Flacon containing the teeth and specimens

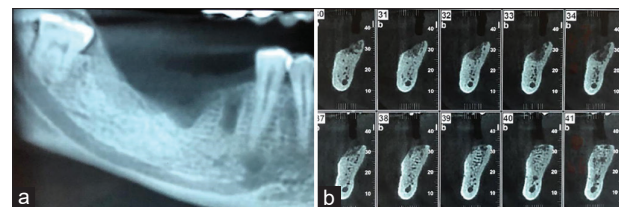


Figure 4: (a) Cropped panoramic reconstruction, 6-month post-surgery showing early signs of bone healing in the extraction sites. The 48 has not been removed. (b) Post-surgery cross sectional cuts showing early signs of bone healing with some remaining low density sites

and that is why we thought about an ameloblastoma since the region, the direction of extension and the age were indicative, but the radiological features were not very indicative since such a growing and an expansile lesion, must, in general, have a multilocular lytic appearance or soap bubble appearance if it was an ameloblastoma.

Treatment of dentigerous cysts can be either surgical removal of the cyst avoiding damage of the concerned tooth or complete enucleation (with concerned tooth/teeth removal) or marsupialization technique,^[9] depending on the size and extent of the cyst, amount of tissue injury, association or not with an erupting tooth, and surgical access.

Marsupialization is a conservative, less aggressive technique than enucleation, normally done in children to avoid the loss of permanent teeth. It consists of creating a window on the cystic wall by incision, partial debridement, suturing the edges of the remaining cyst with the surrounding mucosa, and putting a catheter to create and maintain a continuity between the cyst and oral environment.^[14]

Martin Woods and Peter Reichart, in their book: Maxillofacial surgery, noted an important factor to take into consideration when marsupialization is done: Since only a part of the cystic lining is removed, so only this part is examined histologically; ameloblastomas or even carcinomas developing in cyst linings may be missed.^[15]

In some cases, the process of marsupialization continues, and the lesion resolves completely without further intervention, but many require secondary surgery to remove the residual lining. In fact.

Tainie *et al.*, in a recent article published in 2020, talked about “the two stages treatment strategies of treating large cysts” which consist first of all, in marsupialization to decompress, reduce the lesion volume and therefore, lowering the risk of damaging anatomical structures. Enucleation will be done as a second stage.^[16] From the above description of marsupialization, patient’s cooperation, long-term follow-up, and maintaining a good oral hygiene are important factors for the success of this technique.

In our case, complete enucleation was performed because already the teeth were floating, the patient had a bad overall oral hygiene and in addition to that, in marsupialization, the decrease of intracystic pressure and shrinkage of the cyst will promote bone remodeling and healing. However, in our case, due to the patient’s age, complete bone healing and remodeling cannot be suspected since in adults, turnover is slower than in younger patients.

Recurrence of dentigerous cysts varies between 0 and 62%. In fact, recurrence rate may be affected by many factors such as treatment modality (higher risk with marsupialization than complete enucleation), lesion size, and length of the follow-up period. To avoid the recurrence of dentigerous cysts, most surgeons perform the complete removal of the cyst with marginal extension and well-done curettage of surrounding tissues.^[17]

Conclusion

Although most dentigerous cysts are considered to be developmental in origin, there are some examples that appear to have an inflammatory pathogenesis.

Thus, from the above article and discussion, we can conclude that the correlation between clinical and radiological examination is necessary in such situations to put a temporary diagnosis, but it is the histopathological study that will give us the final and definitive diagnosis which will lead us to the appropriate treatment.

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