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Plexiform ameloblastoma arising from a huge dentigerous cyst – a case report

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Abstract

Dentigerous cysts (DCs) are one of the most common types of cysts occurring in the jaw, which clinically present as an asymptomatic unilocular radiolucency enclosing the crown of an unerupted or impacted tooth. In most cases, the diagnosis of a DC is straightforward; but even radiographically, a typical DC can be diagnosed as something else, such as a dental follicle, an odontogenic keratocyst or a unicystic ameloblastoma on histological analysis. We report a case of an 11-year old boy with an initial diagnosis of a dentigerous cyst, who underwent enucleation under general anaesthesia, and revealed features suggestive of plexiform ameloblastoma on post-operative excisional biopsy.

Keywords

dentigerous cyst; plexiform ameloblastoma; excisional biopsy; enucleation

Introduction

The dentigerous cyst is the most common type of noninflammatory odontogenic cyst and the most common cause of a pericoronal area of lucency associated with an impacted tooth. A dentigerous cyst forms within the lining of the dental follicle when fluid accumulates between the follicular epithelium and the crown of the developing or unerupted tooth. Most DCs manifest in adolescents and young adults and often form around the crown of an unerupted mandibular third molar. Small DCs are usually discovered in radiographic examinations that are taken to investigate other symptoms or a failure of tooth eruption. However, they can grow extremely large asymptomatically and remain undetected until they enlarge enough, causing bone expansion and asymptomatic facial swelling [1]. On radiographic examination, they appear as well-defined, round, corticated, lucent lesions around the crowns of impacted teeth, usually third molars. A differential diagnosis of pericoronal radiolucency should include odontogenic keratocyst, ameloblastoma and other odontogenic tumors. Ameloblastic transformation of a dentigerous cyst lining should also be a part of the differential diagnosis [2]. The histological diagnosis of these lesions is therefore critical. Here, we report a case of an 11-year old boy, who was initially diagnosed as a case of a huge dentigerous cyst on the basis of radiological and cytological examination, underwent enucleation under general anaesthesia, and revealed features of plexiform ameloblastoma on post-operative excisional biopsy.

Case Report

A healthy 11-year old boy presented to our outpatient department with an asymptomatic swelling over the right cheek that had been present for 3 months. His past medical history was unremarkable. An intraoral examination revealed a right mandibular, buccal, cortical expansion extending from the anterior border of the ramus to the second premolar and extending buccally into the cheek, with normal overlying mucosa. Egg shell crackling was observed on palpation of the swelling. There was no nerve deficit or adenopathy in the head and neck region. Panoramic radiography (Figure 1) showed a large, well defined expansile lucent mass in the ramus of right mandible, causing its expansion, which was associated with an unerupted third molar tooth; all four of the patient's wisdom teeth were unerupted.

Contrast- enhanced maxillofacial CT (Figure 2 [a] & [b]) demonstrated a well corticated unilocular lucent lesion measuring 7 x 3.7 x 3.1 cm involving right ramus and angle of mandible, which was completely surrounding the crown of an impacted third molar tooth lying eccentrically.

Fine needle aspiration revealed 20 ml of blood tinged thin fluid; smears prepared showed dense acute and chronic inflammatory cells, few anucleated squames along with cystic macrophages in a blood mixed proteinaceous background, suggestive of a benign inflammatory cystic lesion (Figure 3). On the bases of cytological and radiological features, we made a diagnosis of dentigerous cyst.

We posted the patient for enucleation of the cyst under general anaesthesia. The buccal flap was raised and periosteal dissection was done to expose the ascending ramus and corpus of the mandible (Figure 4). 60-70 ml straw coloured fluid was aspirated from the cyst (Figure 5).

The second molar tooth was extracted for better exposure. The cyst was enucleated along with the associated third molar, and the specimen (Figure 6) was sent for histopathological analysis.

No fracture of the mandible was seen while removal of the cyst. The cavity (Figure 7) was irrigated regularly; healing was uneventful.

On gross examination, the specimen measured 6 x 5 x 3 cm in size. External surface showed areas of hemorrhage and congestion and base of the third molar tooth at one end. On opening the cyst, multiple sessile polypoidal projections were seen ranging in size from 2.5 x 1.5 x 0.5 cm. On microscopy (Figure 8), section from the polypoidal structures showed plexiform arrangement of tumor cells comprising of enamel origin with peripheral ameloblastic columnar cells surrounding areas of edematous stellate reticular like cells. Section from the cyst wall showed odontogenic epithelium with fibrocollagenous stroma along with hemosiderin laden macrophages and foreign body giant cells.

The histological features suggested that the lesion was a dentigerous cyst with ameloblastomatous changes. Due to the pattern of epithelial proliferations, we could suggest that the growth had formed into a plexiform unicystic ameloblastoma. The final histopathological analysis confirmed it to be a plexiform ameloblastoma. No recurrence was noted during the post-operative follow-up of six months.

Discussion

A dentigerous cyst is the most common cause of pericoronal radiolucency which is associated with impacted teeth. Because they are asymptomatic, dentigerous cysts are usually diagnosed on

routine dental radiographs. The diagnosis of a dentigerous cyst is based on a combination of radiographic and histopathological features. Dentigerous cysts form within the lining of the dental follicles when fluid accumulates within the follicular epithelium and the crown of developing or unerupted tooth [1]. Radiographically, the dentigerous cyst typically appears as a well-circumscribed, unilocular, usually symmetric radiolucency around the crown of an impacted tooth [2], as seen in our case too. Dentigerous cysts may enlarge causing extensive bone resorption and even pathological fractures. The greater the size of the cyst, the higher the risk of neurologic damage caused by trauma during and after surgical removal and of mandibular fractures resulting from the postoperative bone defects [3]. However, in our case, in spite of the large size of the cyst, there was neither any neurologic damage nor any pathological fractures.

Most of the dentigerous cysts manifest in the second and third decades of life, with peak incidences in teenages, often developing around the crowns of mandibular third molars as it was seen in our case. An ameloblastoma is a benign and a locally aggressive tumor which arises from the mandible or less commonly, from the maxilla. Robinson and Martinez first described unicystic ameloblastomas which are variants of ameloblastomas, and refer to those cystic lesions that show clinical and radiological characteristics of odontogenic cysts, but which on histological examination, show typical ameloblastomatous epithelium which lines part of the cyst cavity[4]. In a clinico-pathological study done on 57 cases of unicystic ameloblastomas, Ackerson classified this entity into three histological groups: luminal unicystic ameloblastoma, intraluminal / plexiform unicystic ameloblastoma and mural unicystic ameloblastoma.

15 to 20% of all unicystic ameloblastomas form in the wall of dentigerous cysts. Since 1925, many had reported the development of ameloblastomas within the walls of odontogenic cysts, among which the most commonly cited were dentigerous cysts [5]. The immuno-histochemical data on Ki-67 expression in ameloblastomas which arise from dentigerous cysts confirm the hypothesis that ameloblastomas which arise from dentigerous cysts have a biological behaviour which is similar to that of unicystic ameloblastomas [6]. The term, 'plexiform unicystic ameloblastoma' refers to a pattern of epithelial proliferation that has been described in cystic lesions of the jaws. It has been considered as a hyperplastic epithelium, rather than an ameloblastoma by some pathologists, because it does not exhibit histological criteria which were previously accepted for ameloblastomas. Gardner et al., in their article, provided histological evidence that plexiform unicystic ameloblastomas were in fact, variants of conventional unicystic ameloblastomas, by reporting ten cases of unicystic ameloblastomas that exhibited both patterns. Further evidence of the ameloblastomatous nature of plexiform unicystic ameloblastomas is that their biological behaviour, even when this pattern occurs alone, is similar to that of conventional unicystic ameloblastomas [7]. Occurrence of ameloblastomas in children and adolescents who are below 18 years of age is uncommon. Only 14.6% of cases of ameloblastomas were seen in children and adolescents among 206 cases which were evaluated by Lucas [8]. Contrastingly, our patient was only 11 years of age.

In view of the reported ameloblastomatous potential of dentigerous cysts, it is thus important to be able to recognize true ameloblastomatous epithelium from ameloblastoma -like epithelium. In most cases of odontogenic cysts, the presence of an ameloblastomatous epithelial lining in inflamed odontogenic cysts is insufficient to diagnose unicystic ameloblastomas, unless other more diagnostic

features of unicystic ameloblastomas are evident. In such cases, other diagnostic criteria which are included to make a diagnosis of unicystic ameloblastomas, as were described by Vickers and Gorlin, are cysts which are lined by an ameloblastic epithelium, with a tall columnar basal layer, a sub nuclear vacuole, reverse polarity of hyper chromatic nucleus and a thin layer of edematous, degenerating stellate reticulum like cells on surface [9]. In our case, all such histopathological features required to make a definite diagnosis of ameloblastoma were seen.

The recommended treatment for dentigerous cysts is marsupialization, because it is the best way to conserve the permanent tooth [10]. Marsupialization also must first indicate when there is no likelihood of damaging anatomic structures. However, most reports agree that the treatment of choice is enucleation of the cyst followed by extraction of the associated tooth if the tooth involved is particularly a third molar [11]. In addition to this, it must not be forgotten that the major disadvantage of marsupialization is that pathologic tissue is left in situ, without thorough histological examination [10]. Although dentigerous cyst is considered a benign lesion, its epithelial lining has the potential to undergo neoplastic change and development of a squamous cell carcinoma is possible.

In summary, our case is consistent with the recognized association of dentigerous cysts and ameloblastomas with unerupted third molar teeth. The coexistence of these three entities also is in keeping with the hypothesis of ameloblastic transformation of dentigerous cysts. The diagnosis of an odontogenic lesion relies heavily on histopathologic evaluation.

In the present case, making a diagnosis was possible only because histopathological examination of the enucleated material was performed. Thus, a histological examination is the most sensitive tool which can be used for differentiation of dentigerous cysts from unicystic ameloblastomas. The importance of enucleation as the first choice of treatment for large, cystic lesions, instead of conservative procedures like decompression and marsupialization which are used for children is highlighted. Though marsupialization might help in the preservation of vital structures, keeping in view, the potential of more aggressive transformation of the cystic lining, complete removal of the lining by enucleation, with emphasis on possible preservation of vital structures, as in our case, clubbed with a thorough follow-up, is more appropriate.

Figures

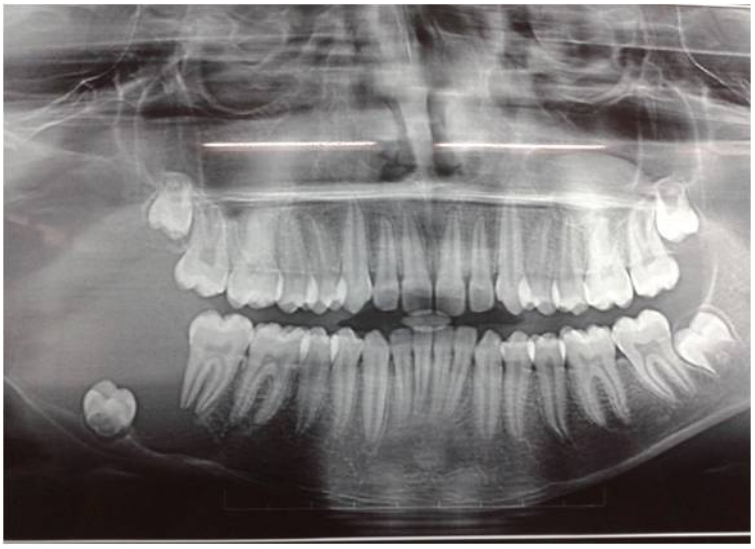

Figure 1: Panoramic radiograph of the huge cyst

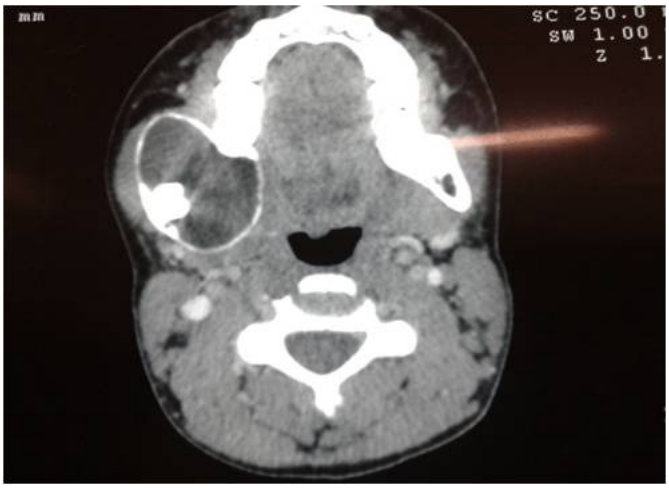

Figure 2A: Axial contrast-enhanced CT image showing the unerupted wisdom tooth centered in the cystic expansile mass

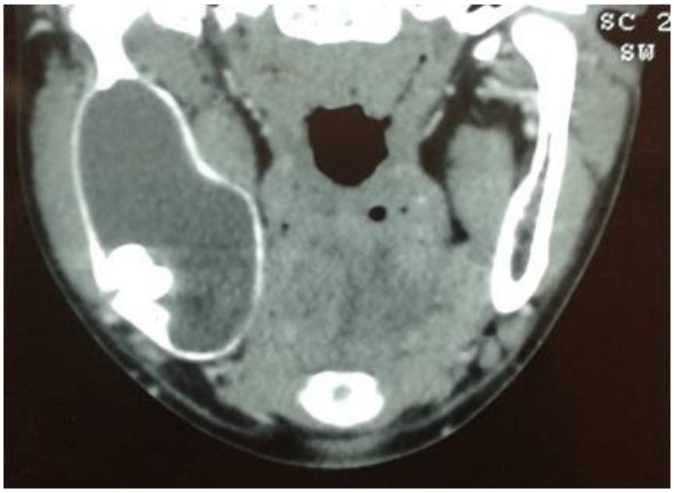

Figure 2B: Coronal contrast-enhanced CT image showing the cyst

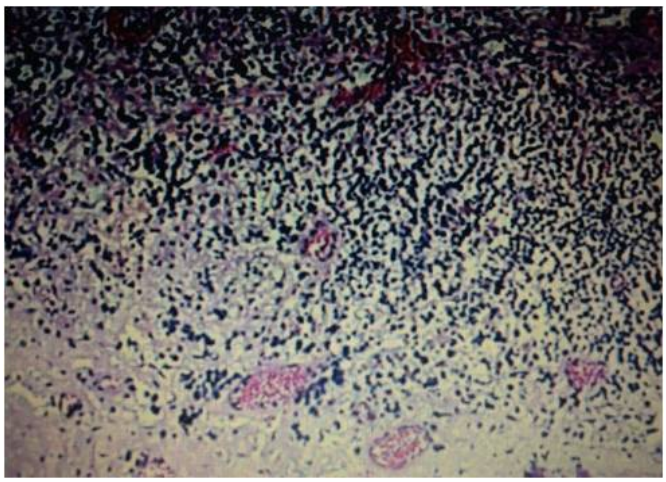

Figure 3: Cytological picture showing features of dentigerous cyst

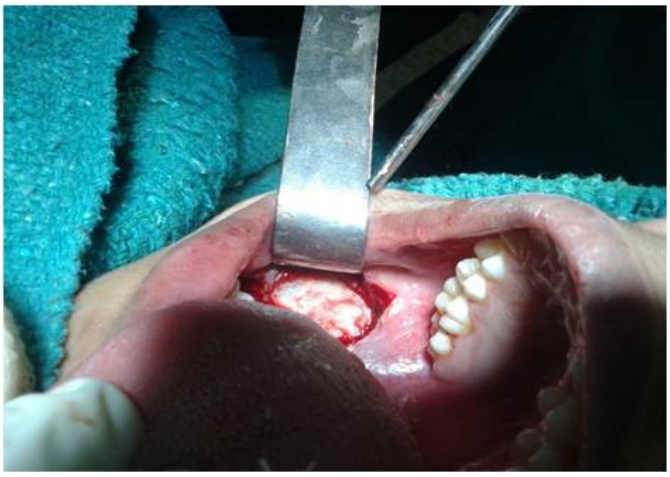

Figure 4: Intraoperative view

Figure 5: Fluid aspirated from the cyst

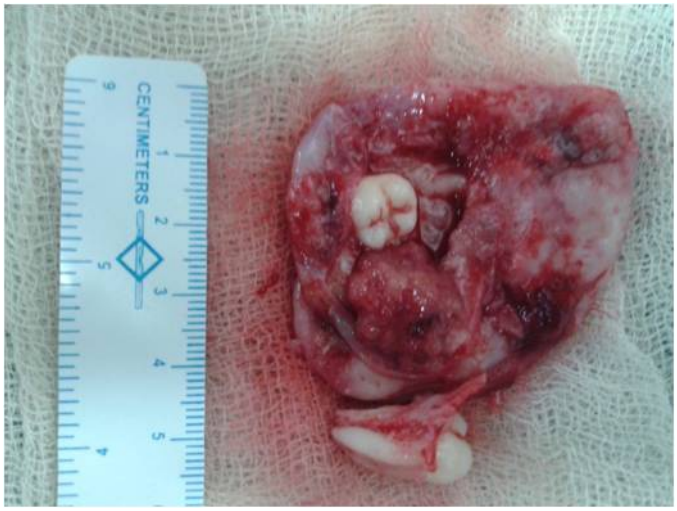

Figure 6: The cyst with the impacted tooth after enucleation

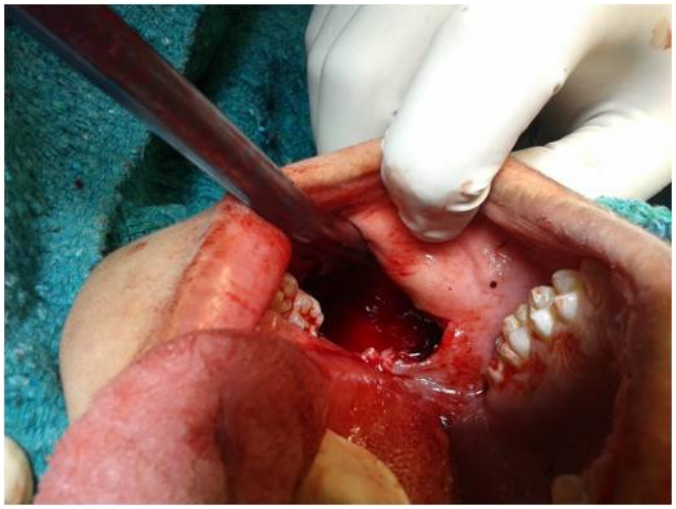

Figure 7: Cystic cavity after enucleation

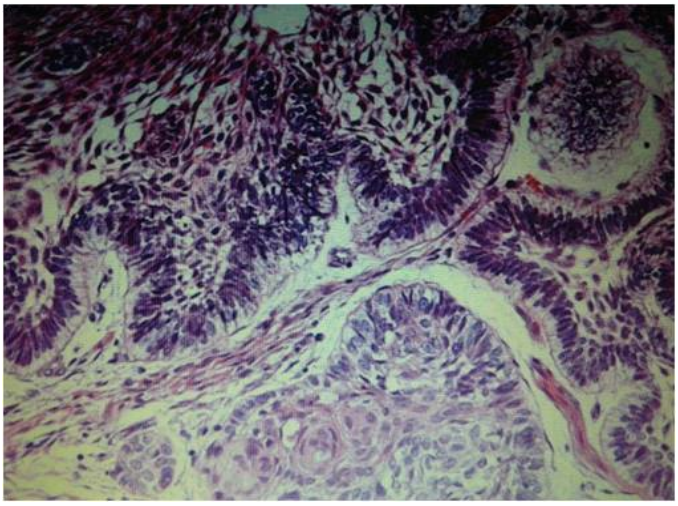

Figure 8: H & E stained photomicrograph (40 X) showing histological features of plexiform ameloblastoma

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