

Rare Case of Huge Bi-Locular Dentigerous Cyst Involving Mandibular Canine

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ABSTRACT

Dentigerous cyst is benign odontogenic cystic lesion that envelops the crown of an impacted tooth. Like odontogenic tumors, dentigerous cysts may grow to large sizes before they are diagnosed, and the large radiolucent cystic lesions associated with an impacted tooth are often diagnosed as odontogenic tumors like unicystic ameloblastoma or others developmental cystic lesions like odontogenic keratocyst, but less likely to be dentigerous cysts. In this case report we highlight the first case of huge bi-locular dentigerous cyst in a 35-year-old woman related with impacted mandibular canine.

Keywords

Dentigerous cyst, Impacted tooth, Odontogenic cysts, Odontogenic tumors.

Introduction

Dentigerous cyst (DC) or follicular cyst is benign odontogenic cystic lesion that envelops the crown of an impacted tooth formed by extension of its follicle because of aggregation of fluid between Nasmyth's membrane and the surface of enamel, and the expansion of this cyst is related to an increase in cyst fluid osmolality and the release of bone resorption factors [1-3]. This eventually leads to the formation of the cyst in which the crown is located within the lumen and roots outside [4].

DCs are more than twice as common in males as females, and two-thirds develop on lower third molars and most of these cysts occur during the second and third decades [1]. Generally, DCs are asymptomatic lesions and are observed during routine radiographic investigations [5,6]. Huge DCs may cause expansion of the cortical bone and resorption of adjoining teeth, but such cases still unfamiliar [7].

The radiographs show a unilocular radiolucent cystic lesion covering the crown of an impacted tooth (central location), and on the side of the tooth (lateral location), or completely around the tooth (circumferential location). The radiolucency area has a well-defined and often sclerotic border [1]. Large DCs can have a multilocular appearance on radiograph, given the existence of bone trabeculae within the radiolucency. However, they are histologically a unilocular lesion [7].

Herein, we reported a rare case of huge bi-locular DC involving permanent mandibular left canine in a female patient. After verifying all the anterior mandibular cases in the literature, we found that this is the first case of bi-locular DC in anterior mandible.

Case Report

A 35-year-old woman consulted the department of oral and maxillofacial surgery because of a mild swelling at the anterior mandible.

History of present illness

The patient complained of a 2-month history of progressive nonpainful swelling of her anterior mandible. The patient denied

any history of pain, fever, purulence or trismus, and she did not report any neurosensory changes.

Clinical examination

The patient had slight lower facial swelling; the mass was hard, non-fluctuant and non-tender to palpation. There were no facial or trigeminal nerve deficits. Neck: there were no palpable mass and no cervical or submandibular lymphadenopathy.

Intraoral

Occlusion was stable and there was no movement in teeth, the interincisal opening was within normal limits. There was buccal expansion of the anterior mandible extending from right mandibular first premolar toward the left mandibular first premolar. All teeth were vital according to electrical test. Blood supply and innervation have been maintained in remaining bone tissue.

Radiographic examination (OPG and CBCT)

There was large bi-locular radiolucent lesion with sclerotic well-defined borders and associated with impacted mandibular left canine. This large lesion extended from left mandibular premolars to right ones, and there were clear signs of destruction of buccal bone lamina (Figures 1 and 2).

On these clinical and radiographic characteristics, differential diagnosis included dentigerous cyst, odontogenic Keratocyst and unicystic ameloblastoma.

Surgical procedure

After the nasal and endotracheal intubation, injection was done in the working area by lidocaine with vasoconstrictor above the periosteum in the lower anterior vestibular region. Then a flap was designed on the teeth adjacent to the cystic lesion (33, 32, 31, 41, 42, and 44) with two release incisions, after that a full thickness mucoperiosteal flap was elevated and the both mental foramina with mental nerves were isolated. After that, the whole cystic lesion was enucleated and the impacted canine was extracted. Carnoy's solution was used until reaching a healthy bone. Finally, Gel foam was applied then the flap was repositioned. VICRYL thread (4-0) was used for sutured (Figures 3, A-B-C-D).

Histologic examination: the microscopic revealed that hyperplastic (about 8 to 20 cuboidal cells in thickness) non-keratinized stratified squamous odontogenic epithelium surrounding the lumen of this cystic lesion. The connective tissue showed interweaving fibers of collagen, vascularity and fibroblasts. Severe cholesterol clefts and lymphocytes infiltration were also seen in the connective tissue, revealing of inflamed DC (Figures 4, A-B-C).

Radiological signs of good bone formation after 6 months including mixed radiolucent - radiopaque area in anterior mandible (Figure 5).



Figure 1: Orthopantomogram (OPG) showing the large bi-locular radiolucent lesion associated with impacted mandibular left canine.



Figure 2: CBCT finding: three sagittal sections of cone-beam computed tomography (CBCT) show: (A) the buccal cortical defect on the first locusus (arrow). (B) The bony septa between two loculi which was lingually from impacted canine (solid arrowhead). (C) Shows the second locusus (open arrowhead).

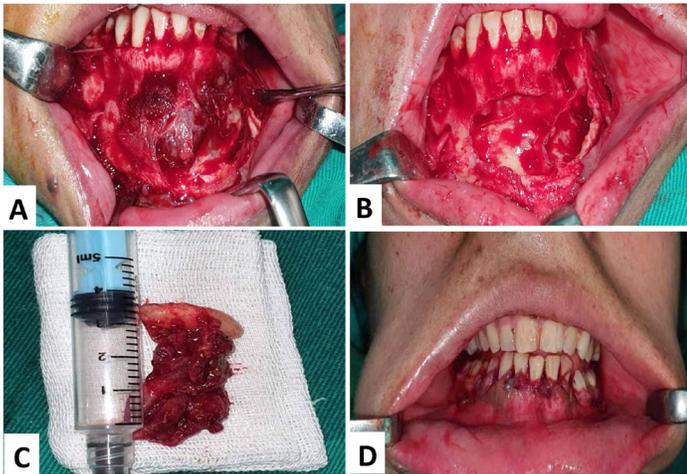


Figure 3: (A) Intra-oral photograph showing the lesion border with evident bone destruction. (B) Intra-oral photograph showing the healthy bone after removal of whole lesion. (C) Post-operative photograph showing the complete enucleation. (D) Post-operative photograph showing the wound suture.

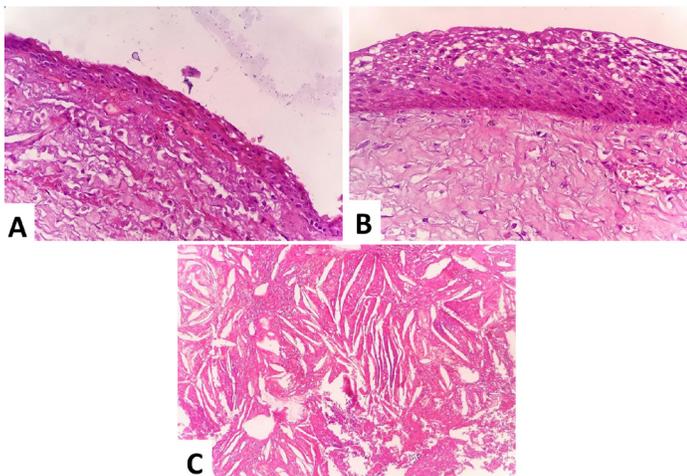


Figure 4: Histological photomicrographs showing: (A) The fibrous connective tissue lined with odontogenic epithelium (H&E, 40 \times). (B) The hyperplastic odontogenic epithelium: 20 layers of cuboidal cells (H&E, 40 \times). (C) The severe cholesterol clefts (H&E, 10 \times).



Figure 5: Orthopantomogram (OPG) showing follow up after 6 months.

This study was legally authorized by the Ethics Committee at General Assembly of Damascus Hospital (Approval ref: 200164), and the signed informed consent for publication was obtained from the patient.

Discussion

DC (sometimes called follicular cyst) is a developmental odontogenic cyst that envelops the crown of impacted tooth, which may be part of the regular dentition or a supernumerary tooth [5]. This cystic lesion arises from the epithelium of the dental follicle and remains attached to the neck of the tooth, enclosing the crown within the cavity of cyst [1,5].

DC may be observed as an incidental radiographic finding or by investigating a clinical expansion, and the potential neoplastic transformation include unicystic ameloblastoma, and carcinomatous transformation include intraosseous squamous cell carcinoma which may derived from changes of the epithelial cells of DC [8-10]. Therefore, follow-up is essential to avoid complications and to get better treatment according to the diagnosis of each different case. Hence, early and correct diagnosis will increase the percentage of success [11].

In addition, it has been suggested that the potential for development of the rare intraosseous mucoepidermoid carcinoma (MEC) may arise from the presence of mucous cells in the DC epithelial lining [12-14].

Some DCs may appear as a multilocular appearance on radiographs because of protruding bony septa from the walls, but this appearance should raise suspicion for odontogenic neoplasms or non-odontogenic lesions [15].

Conclusions

Huge DCs are unfamiliar, and such benign lesions related with impacted tooth may be odontogenic tumors. Therefore, it is important that the final diagnosis decision be based on histopathological examination, rather than radiographical finding only.

Hence, early diagnosis and proper treatment planning for such uncommon cases is necessary to avoid further complication, and a long-term follow up is essential in these cases.

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