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CASE REPORT

Odontogenic keratocyst in the maxillary sinus: Report of two cases

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Summary The odontogenic keratocyst (OKC) is well known for its tendency to recur, potential aggressive behaviour and defined histopathological feature. OKC occurrence in the maxilla is unusual and its appearance in the maxillary sinus very uncommon. This article reports two distinct cases of OKCs associated with unerupted molars in the maxillary sinus of two boys. The lesions were surgically treated and no recurrence has been observed on follow-up. OKC clinical features and treatment are discussed.

Introduction

The odontogenic keratocyst (OKC) is a distinct entity from the odontogenic cysts that deserves special attention due to its aggressive clinical behaviour and high rate of recurrence. Multiples OKCs may be associated to basal cell nevus syndrome. Due to unspecific clinical and radiographic features, it may be confused as ordinary cysts, leading to underdiagnosis and undertreatment, resulting in unnecessary recurrences. Successful treatment depends on the precise diagnosis, adequate surgical procedure and follow-up.[1,2]

The aim of this paper is to report two cases of OKCs located in the maxillary sinus. Differently from the other rare reports presented in the literature concerning OKC in the maxillary sinus,[3,4] in our cases the lesions appeared totally limited in the sinus cavity, without involvement of the maxillary alveolar bone. They were surgically removed and the patients have been followed for eight and five years, respectively, showing no signs of recurrence.

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Case reports

Case 1

A 17-year-old Caucasian boy presented with complaint of a persistent headache, present for a week. The previous medical history was not contributory and no alteration was developed by extra and intra-oral examination. Lateral and post-anterior radiographs showed a discrete opaque mass with an image of the third molar in the left maxillary sinus. The lesion seemed to be totally confined into the left maxillary sinus cavity, without nasal sinus or maxillary alveolar bone involvement (Fig. 1(A)). Under local anesthesia, an access to maxillary sinus lateral wall through canine depression was prepared. After osseous trepanation, a cystic lesion could be visualized. An aspiration biopsy exposed milky white liquid, suggestive from keratocyst (Fig. 1(B)). The cyst containing the tooth was enucleated in pieces and soft tissue curettage was executed. The histopathological exam confirmed diagnosis of odontogenic keratocyst (Fig. 1(C)). The patient has been followed-up for eight years and shows no signs of recurrence.

Case 2

A 14-year-old Caucasian boy was referred with chief complaint of bad taste in mouth, persistent for 2 weeks. Intra-oral clinical examination revealed a purulent fistula in the buccal face, above the second right upper molar. Past medical history was unremarkable. Computed tomography and lateral radiograph showed an ectopic second right upper molar involved by a radiopacity filling in the upper posterior portion of the maxillary sinus (Fig. 2(A)). No alveolar bone involvement was noticed. Under general anesthesia, a Caldwell-Luc approach was performed and the cystic lesion could be removed in pieces (Fig. 2(B)). Curettage was carried out in the sinus walls. Histological findings established diagnosis of OKC (Fig. 2(C)). The patient has been followed-up for five years and is disease-free.

Discussion

Since the recognition of the odontogenic keratocyst,[5] and later having its histological criteria defined,[6] this lesion has been theme of investigation and study motivated by

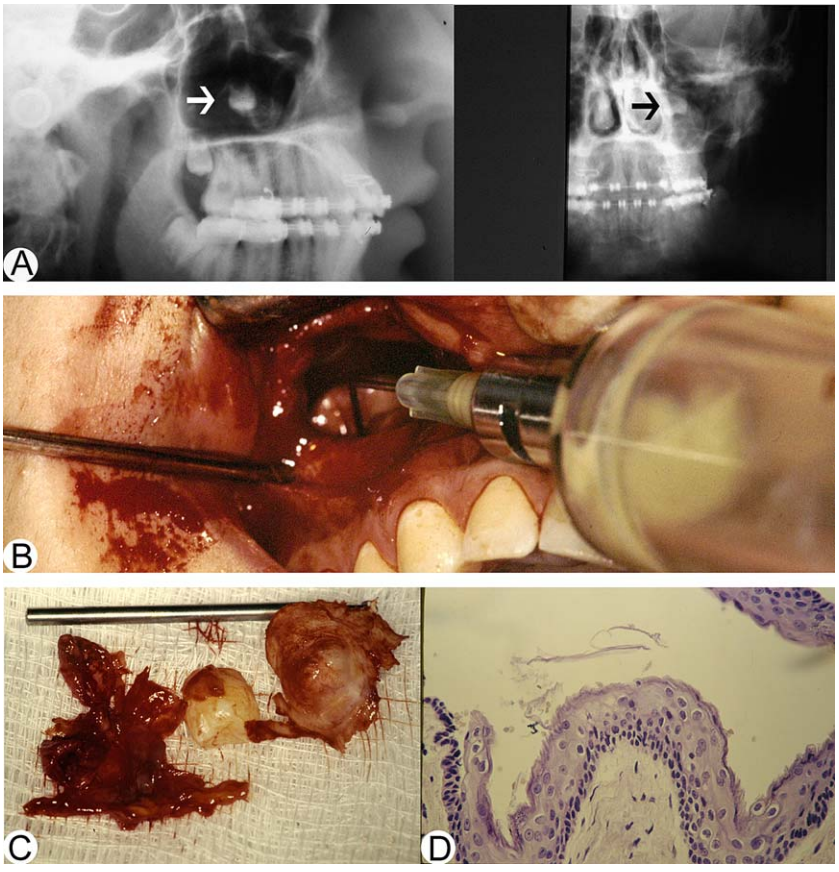

Figure 1 (A) Lateral and post-anterior radiograph showing unerupted tooth in the left maxillary sinus surrounded by a discrete opaque mass (arrows). (B) Aspirating biopsy of the cystic lesion showing milky liquid. Patient presented without brackets because he had finished his orthodontic treatment before surgery. (C) Removed tooth with fragmented cystic lesion. (D) Light microscopic image of the fragmented cyst showing epithelial lining exhibiting parakeratotic layer, thin spinous cell layer and hyperchromatic columnars cells in the basal layer, with no ridges into the connective tissue (H&E 40×).

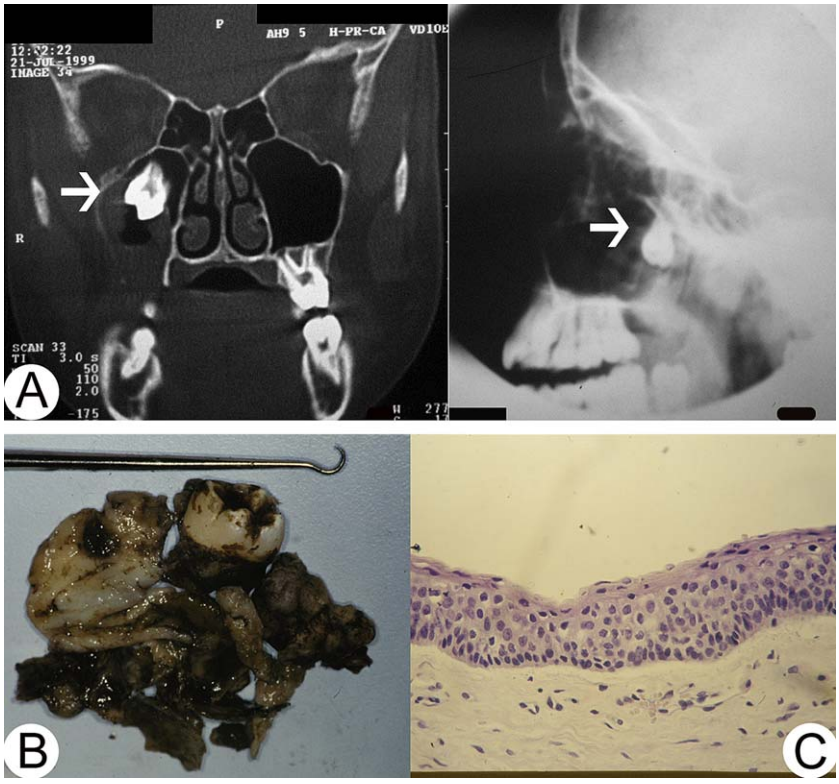

Figure 2 (A) Computed tomography scan and lateral radiograph showing unerupted tooth associated with an opaque filling in superior portion of the right maxillary sinus (arrows). (B) Removed tooth with fragmented cystic lesion (formalin-fixed). (C) Micrograph showing epithelial lining with parakeratotic layer, about six layers of spinous cells and hyperchromatic columnars cells in the basal layer in a palisaded arrangement. The epithelial-connective tissue junction is plain (H&E 100×).

its tendency of recurrence and potential aggressive nature. It was suggested that OKC originates from dental lamina remains.[7] Some authors support that it should be considered a benign cystic neoplasm,[8–10] due to its growth capacity and development characteristics related to the mutation of a suppressor tumor gene, PTCH, found in sporadic and in associated to basal cell nevus syndrome keratocysts.[11] Recently, intracystic fluid pressure was found to be involved in OKC growth.[12]

The OKC has a predilection for men, occurs significantly more in the posterior region of the mandibule and affect more people from second and third decades of life.[1,2,8,13–16] It seems that less than 1% of all cases of OKC occur in the maxilla with sinus involvement.[2,14] One interesting aspect in our presented cases is that the OKCs were completely restricted in the maxillary sinus without alveolar bone or erupted teeth association, while the previous cases reported large OKCs that probably developed in the maxillary bone and expanded to the sinus.[3,4]

Recurrence is usual,[1,2,6,8,13–18] occurring principally in the first 5 years after operation,[2,15,16,18,19] but may occur in much longer time intervals. Recurrence rate was found to vary from 0% to about 62%, depending on the kind of treatment management and follow-up period.[13–18] The recurrence of OKC is thought to be based on great mitotic activity and growth potential found in epithelium, further than other sources of recurrences such as remnants of dental lamina and epithelial islands. These findings lead to surgery recommendation to eradicate epithelium components of these cysts and excision of the mucosa where OKC adheres.[8]

Various treatment alternatives based on surgical approaches have been suggested, such as marsupialization, enucleation, enucleation with Carnoy's solution, enucleation with cryotherapy, curettage and resection.[13–18] Simple enucleation was associated to a higher recurrence rate,[13–18] while resection[17] and enucleation with bone curettage presented lower rates.[16] Special attention should be given to the dentate area if the enucleation is chosen as treatment, due to higher rates of recurrence found in OKC associated with teeth.[13] In our exhibited cases treated with enucleation and curettage, no recurrence was found after 8 and 5 years of follow-up, which was expected, because cysts were not adhered to bone.

As OKC appearance in the maxillary sinus is rare, its radiographic image in such situation may be misinterpreted. Computed tomography can provide information on the extent of these lesions, contributing to diagnosis and preoperative preparation. OKC has clinical diagnostic difficulties due to relative lack of specific clinical and radiographic characteristics.[18] Because OKC has features similar to dentigerous cyst (radiographic image) and ameloblastoma (mean age at diagnosis, mandibule predilection, propensity to recur and radiographic appearance), and these are the most common provisional diagnoses for OKC,[2] ideally a biopsy specimen examination and accurate clinical, radiographic, trans-surgical observation are essential to determine the most effective treatment in order to avoid recurrence.[17,19]

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