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## CASE REPORT

# Odontogenic keratocyst in the maxillary sinus: Report of two cases

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### KEYWORDS

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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.

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## 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. Multiple 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.<sup>1,2</sup>

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,<sup>3,4</sup> 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

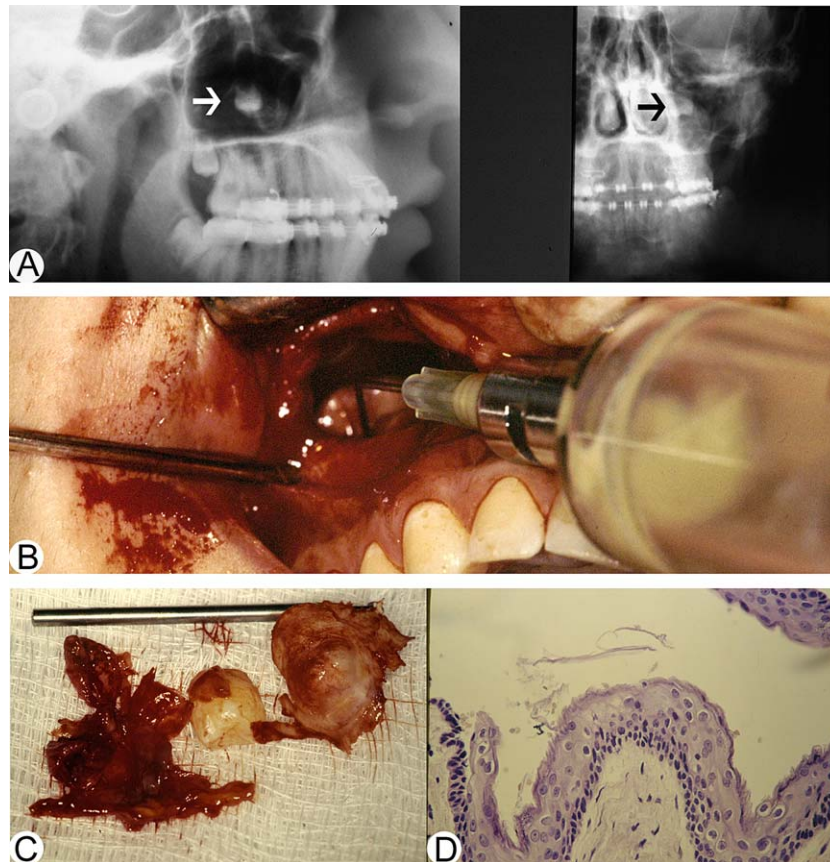
A 17-year-old Caucasian boy presented with complaint of a persistent headache, present for a week. The previous medical history was not contributory and no alteration was developed by extra and intra-oral examination. Lateral and post-anterior radiographs showed a discrete opaque mass with an image of 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,<sup>5</sup> and later having its histological criteria defined,<sup>6</sup> 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 columnar cells in the basal layer, with no ridges into the connective tissue (H&E 40 $\times$ ).

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