## CASE REPORT

# Odontogenic keratocyst in maxillary sinus with invasive behaviour

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Odontogenic keratocyst is a cystic lesion characterized by a high rate of recurrence. This report describes a rare case of ciliated epithelium-lined odontogenic keratocyst in the maxilla of a 27-year-old female. Panoramic radiography showed a lytic lesion on the right maxilla associated with an impacted molar tooth. Computerized tomography image revealed the involvement of the lesion with the right maxillary sinus, destroying the sinus floor. Histopathologically, the typical keratinized epithelial-lined cyst of odontogenic keratocyst abruptly changed into a ciliated epithelium, suggesting the fusion of both these epithelia rather a metaplastic transformation. The biological behaviour of odontogenic keratocysts is discussed.

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#### **Case description**

A 27-year-old woman complained of tooth pain in the right first maxillary molar. Intra-orally the right maxillary second molar was absent. Computerized tomography (CT) scans showed a lesion in the right maxilla extending to the maxillary sinus floor. An impacted right second maxillary molar was involved in the lesion (Fig. 1). A CT axial view disclosed an opaque lesion in the right maxillary sinus, partially filling the right maxillary sinus and destroying the right sinus floor (Fig. 2). The patient underwent a resection by curettage and no evidence of recurrence was registered 5 years later. Microscopically, the cyst was covered with a stratified squamous epithelium with few layers, a parakeratinized, corrugated surface, and a palisading pattern of basal cells with polarized and hyperchromatic nuclei. The cystic lining lost its stratified aspect and abruptly became columnar and ciliated (Fig. 3). Two connection points between the



Figure 1 A computerized tomography scan showing a lesion in the right maxilla extending to the maxillary sinus floor. Note the involvement of the right maxillary second molar impacted tooth.



**Figure 2** Computerized tomographic axial view showing a lesion involving the right maxillary sinus with cloudy appearance and destroying the lateral wall of the maxillary sinus.

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**Figure 3** Typical odontogenic keratocyst lining with a corrugated parakeratinized stratified squamous epithelium and palisading arrangement of basal cells. Note the abrupt change of cystic lining (haematoxylin and eosin).



Figure 4 High magnification of connection points of both epithelia (haematoxylin and eosin).

respiratory epithelium and odontogenic keratocyst lining were observed (Fig. 4). Daughter cysts in the cystic wall were also present (Fig. 5).

#### Comments

Philipsen (1), in 1956, described an odontogenic cyst with keratinous epithelial lining and some typical characteristics pointed out by Pindborg and Hansen (2), such as uniform thickness, a keratinized pattern with a corrugated surface, palisading of basal cells with hyperchromatic nuclei and a daughter cyst in the cystic wall. Unlike other odontogenic cysts, the odontogenic keratocyst has local aggressive behaviour producing a high rate of recurrence up to 62.5% (3). Immunohistochemical studies suggest the hypothesis that



**Figure 5** Daughter cysts in the cyst wall and separation areas between epithelium lining and underlying connective tissue (haematoxylin and eosin).

odontogenic keratocysts represent a low-grade neoplasm (4–6). Furthermore, genomic studies of odontogenic keratocysts reveal DNA mutation, supporting the neoplastic theory, rather than a developmental origin (7).

The maxilla is involved in 23.5% of odontogenic keratocysts (3). Two cases of ciliated epithelium in odontogenic keratocysts in the maxilla have been reported. Although the origin of this epithelium remains unknown, it has been suggested that it is a true metaplasia (8, 9). In the present study, a case of odontogenic keratocyst in the maxilla partially lined with ciliated epithelial changes is presented. A CT examination disclosed a lesion in the right maxilla invading the sinus floor. The microscopic examination revealed a cyst covered by two different epithelia. The typical keratinized epithelium of odontogenic keratocyst changed abruptly to a respiratory ciliated lining, suggesting a replacement mechanism rather than a true metaplasia. It is reasonable to consider that this microscopic characteristic suggests an aggressive biological behaviour of the odontogenic keratocyst. Further molecular studies will be useful in providing a clearer understanding of the biological behaviour of odontogenic keratocysts.

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