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Case report

A case of an epidermoid cyst arising in the maxillary sinus

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ABSTRACT

In the oral and maxillofacial regions, epidermoid cysts usually develop in the floor of the mouth and rarely in other sites. We describe a case of an epidermoid cyst arising in the right maxillary sinus. A 27-year-old man was referred to our department for swelling in the right buccal region. Computed tomography showed a dense soft-tissue mass that extended widely into the maxillary sinus, resulting in thinning of the cortical bone of the exterior wall of the maxillary sinus. The cyst was enucleated via the intraoral approach with the patient under general anesthesia. Histopathologically, the wall was lined with a thin layer of keratinizing squamous epithelium and fibroma connective tissue with no skin appendages. The diagnosis was an epidermoid cyst. There has been no evidence of recurrence during the 3-year follow-up.

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1. Introduction

Epidermoid and dermoid cysts are developmental lesions containing elements of the ectoderm and mesoderm. The cysts are believed to arise from the inclusion of ectodermal elements within the neural groove at the time of closure at 3–4 weeks of embryonic life [1]. About 7% of the cysts develops in the head and neck region, and only 1.6% develops in the oral cavity [2]. Most often they are located in the submental region and floor of the mouth. Other common locations are the lateral tongue, lateral pharyngeal wall, soft palate, and within the jaw [3].

We report a rare case of an epidermoid cyst arising in the right maxillary sinus, present a review of the literature, and discuss the histogenesis of the lesion.

2. Case report

A 27-year-old man was referred to Chiba University Hospital in September 2008. The patient noticed swelling in the right buccal region about 1 month previously (Fig. 1). He had bilateral chronic maxillary sinusitis with nasal obstruction during

childhood. Whereas the patient used medication occasionally, he did not undergo maxillary sinus surgery. An intraoral examination showed non-tender swelling from the upper right first molar region to the maxillary tuberosity corresponding to the buccal region. A panoramic radiograph showed a well-circumscribed, dome-shaped, radiopaque lesion at the inferior aspect of the right maxillary sinus (Fig. 2). Computed tomography (CT) and magnetic resonance imaging (MRI) showed a cystic lesion about 40 mm × 40 mm × 36 mm in the right maxillary sinus extending into the oral cavity. The lesion had expanded and displaced the roof and the lateral wall of the right maxillary sinus (Figs. 3 and 4). The patient had no ophthalmic symptoms.

A biopsy was performed and a full-thickness mucoperiosteal flap was elevated to show thin alveolar bone with a perforation at the crest. Microscopically, the tissue was comprised of a cyst lined with orthokeratinized squamous epithelium supported by dense fibrovascular connective tissue. No cells suggested the presence of a malignancy. The lesion was diagnosed as a dermoid or epidermoid cyst.

With the patient under general anesthesia, we surgically removed the cyst via the intraoral approach in October 2008. The cyst was removed as en bloc from a bone defect of maxillary sinus (Fig. 5). The cyst was filled with necrotic keratinaceous detritus. Microscopically, the cyst was lined with thin stratified squamous epithelium. No evidence of dermal appendages was present in the cystic wall. A chronic inflammatory infiltrate was seen in the connective tissue (Fig. 6). Based on these findings, the lesion was diagnosed as an epidermoid cyst.

The postoperative course was uneventful, with no signs of recurrence during the follow-up period of 3 years after enucleation.

[☆] AsianAOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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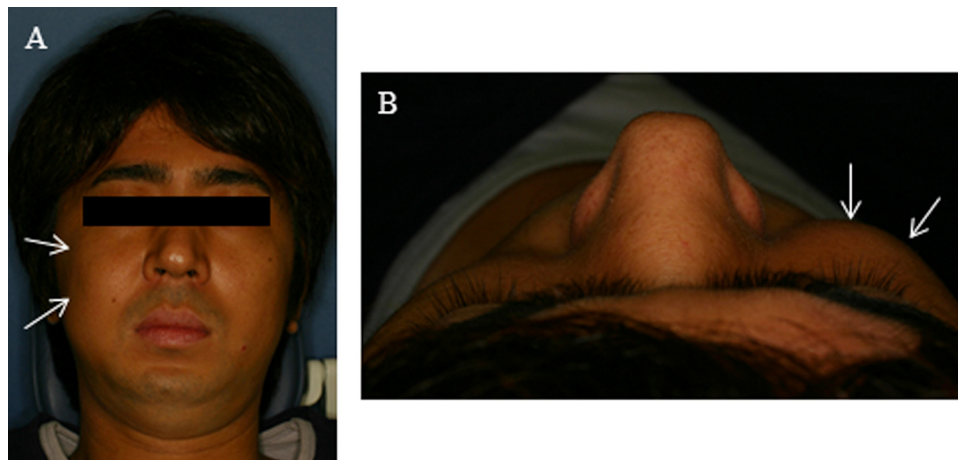


Fig. 1. Clinical photographs show (A) a frontal view and (B) swelling on the right side of the face.



Fig. 2. A panoramic radiograph shows a dome-shaped radiopacity in the right maxillary sinus.

3. Discussion

Mayer updated the concept of dermoid cyst to describe three histologic variants: a true dermoid cyst, an epidermoid cyst, and a teratoid cyst [4]. The true dermoid cyst is characterized by epithelial-lined cavities with keratinization and identifiable skin appendages. An epidermoid cyst is lined with simple squamous epithelium with a fibrous wall and no adnexal structures and is similar to a cholesteatoma, as reported in the otorhinolaryngology,

embryologically, and pathologically literature [5]. A teratoid cyst, also known as a complex cyst, is lined with different types of epithelia, from simple squamous epithelium to the ciliated respiratory type, containing derivatives of ectoderm, mesoderm, and endoderm [4]. Thus, all dermoid cysts have an epithelial lining, but epidermoid cysts do not contain adenexa.

An epidermoid cyst can arise anywhere in the body. The common locations in the head and neck region are the submental region, floor of the mouth, lateral tongue, lateral pharyngeal wall, soft palate, and within the jaw [3,6]. Therefore, the maxillary sinus is an uncommon place for an epidermoid cyst to develop.

The most frequent congenital cause of an epidermoid cyst is aberrant ectoderm in the area of the branchial arch and groove at the age of 3–4 weeks. In acquired elements, aberrant epithelial components in deep tissue resulting from trauma, surgical complications, and inflammation are hypothesized mechanisms of onset [7]. Torske et al. suggested that embryonic tissue trapped in the nasal cavity might have migrated into the developing maxillary sinus, eventually giving rise to a cyst [8]. In the current case, there was no history of trauma or a maxillary sinus surgery. It had been suggested that chronic sinusitis from childhood may be a cause of this disease.

Most patients with epidermoid cysts are from 10 to 35 years of age. Vargas Fernandez et al. proposed that cystic growth may be constrained by hormonal stimulus during puberty, which would explain the greater incidence in young adults [9].

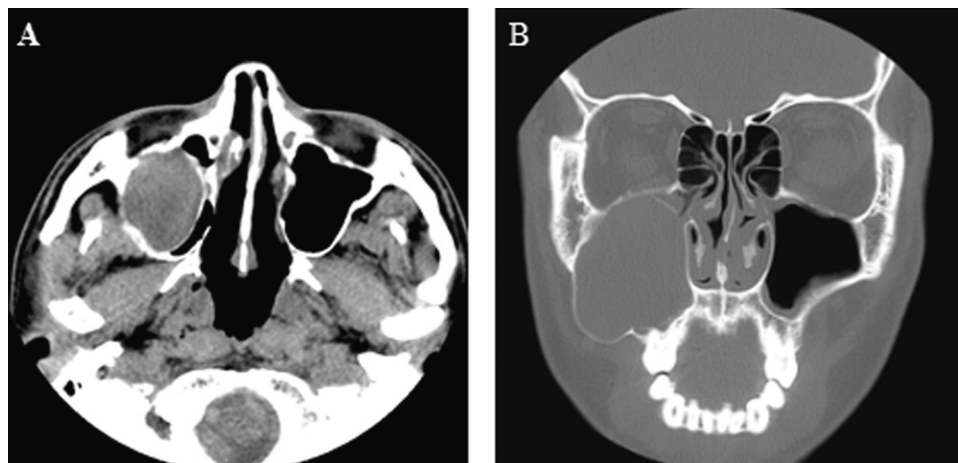


Fig. 3. Computed tomography shows the cyst in the right maxillary sinus: (A) an axial section at the level of the maxillary sinus area and (B) a coronal section at the upper first molar region.

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