

Case Report

Surgical ciliated cyst of the maxilla: a rare pathology of the maxillary sinus

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Abstract Surgical ciliated cyst of the maxilla is a rare complication following surgical procedures or trauma involving the maxillary sinus. It can occur at any time after any procedures or trauma involving the maxillary sinus even though many years have lapsed. Clinically it may mimic other cysts of the maxillary sinus therefore a thorough history taking is necessary for diagnosis. Treatment is usually by enucleation or marsupialisation. This report highlighted a case of surgical ciliated cyst involving the right maxillary sinus probably secondary to maxillary sinus procedures, which were performed 30 years prior to presentation.

Keywords: maxilla, maxillary sinus, surgical ciliated cyst.

Introduction

Surgical ciliated cyst of the maxilla is a benign cyst of the maxillary sinus that formed after a period of time following surgery or trauma involving the Schneiderian membrane. It is also known as postoperative maxillary cyst, paranasal cyst or respiratory implantation cyst. Many of the cases were reported in the Japanese literature and it occurs in about 20% of patients who underwent surgery involving the maxillary sinus. Prevalence outside Japan is relatively low which explained the very low number of English-language publications on this pathology. A literature search using Pubmed revealed only 18 English-language articles from the year 1958 to 2016, reporting 21 cases of surgical ciliated cyst in the maxillo-mandibular region. This finding gave rise to the hypothesis that geographical factor may play a role in the development of this cyst (Kaneshiro *et al.*, 1981, Nishioka *et al.*, 2005). Men are affected slightly more than women. The highest incidence is between the fourth and sixth decades of life (Bulut *et al.*, 2010). Most cases were located in the molar and premolar regions due to their proximity to the maxillary sinus. Diagnosis of such rare case poses a

great challenge due to its similarity to other cyst or tumour of maxillary sinus.

Case report

A 43-year-old man was referred to the Oral and Maxillofacial Surgery Clinic, Faculty of Dentistry, University of Malaya by an otorhinolaryngologist suspecting of ameloblastoma of the right maxilla. The patient presented with a diffuse swelling over the right maxillary region. The swelling was first noticed about one year ago. He also complained of suffering from blocked right nostril. He has had maxillary sinus surgeries done twice by another otorhinolaryngologist about 30 years prior to presentation to manage his chronic maxillary sinusitis. His age was between 13 and 15 years when this happened.

At clinical examination, the overlying skin of the swelling was observed to be of normal skin colour, and the swelling was slightly tender on palpation but not pulsating and there was no numbness over the infraorbital nerve distribution area. Intraorally, the upper right buccal vestibule was obliterated with expansion of the alveolar segment over the molar region. Teeth 14, 15, 16 and 17 were not tender to

percussion. Periodontal pocketing was not present in all the four teeth and surrounding gingivae were of normal colour and appearance. Tooth 15 was root-treated. Other teeth were otherwise vital. Pre-operative computed tomographic (CT) scan showed an extensive radiolucent multilocular cystic lesion within the right maxillary sinus measuring 35 mm x 31 mm x 34 mm, which obliterated the right osteomeatal complex and resorbed both anterior and posterior wall of the maxillary sinus. However, the floor of the orbit was spared (Fig. 1). Straw-coloured fluid was aspirated from the lesion. An incisional biopsy was performed through the buccal vestibule and the result suggested the presence of a surgical ciliated cyst of right maxillary sinus. A decision was made to perform enucleation of the cyst under general anaesthesia.

Intra-operatively, a submarginal flap was raised in order to avoid possibility of recession of the involved teeth. The cyst was enucleated in total together with the sinus lining (Fig. 2). The bony cavity was

packed with ribbon gauze soaked with Whitehead varnish, which helped in preventing haematoma formation within the maxillary sinus, which may lead to infection. The wound was then closed primarily. Post operatively patient was given diclofenac sodium 50 mg every 8 hour and amoxicillin/clavulanic acid 625 mg every 12 hour for one week. Healing was uneventful and the Whitehead varnish ribbon gauze was removed 2 weeks after surgery. Histopathological examination of surgical specimen showed a cyst lined by thin epithelium composed mainly of pseudostratified ciliated columnar type, which confirmed the diagnosis of a surgical ciliated cyst of the right maxillary sinus (Fig. 3). Patient was reviewed on a regular basis. A cone-beam computed tomography (CBCT) scan was taken at 9 months post-operative and compared to the pre-operative CT scan. There was no abnormality detected both clinically and radiographically during the review visit.

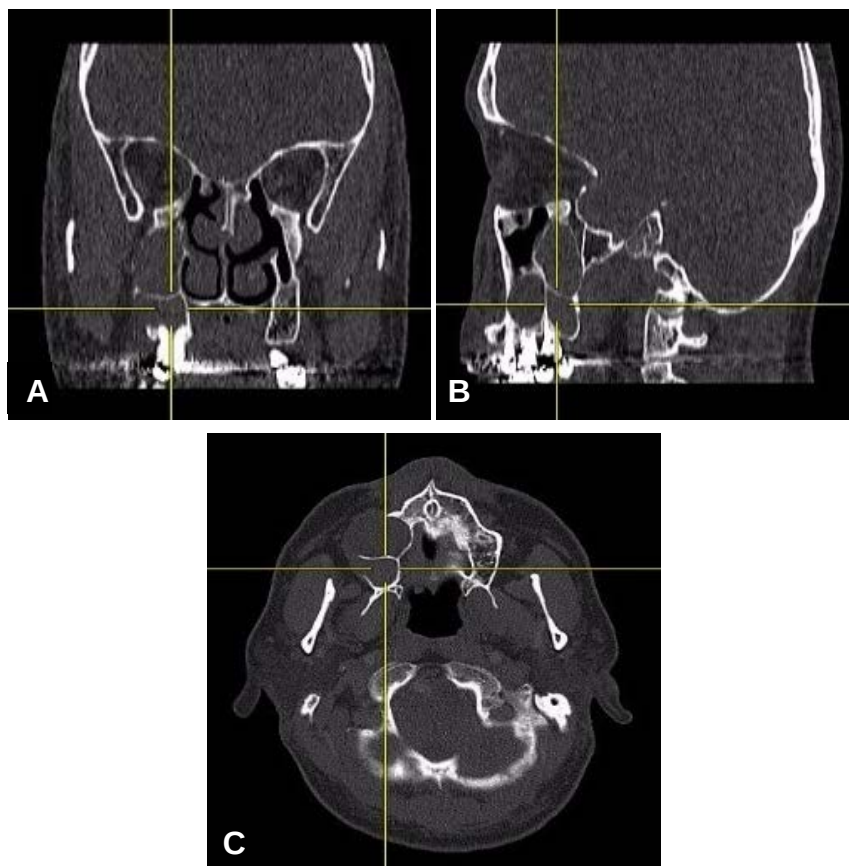


Fig. 1 Pre-operative CT scans showing the location of the cyst (crossing lines). (A) coronal view, (B) sagittal view and (C) axial view).

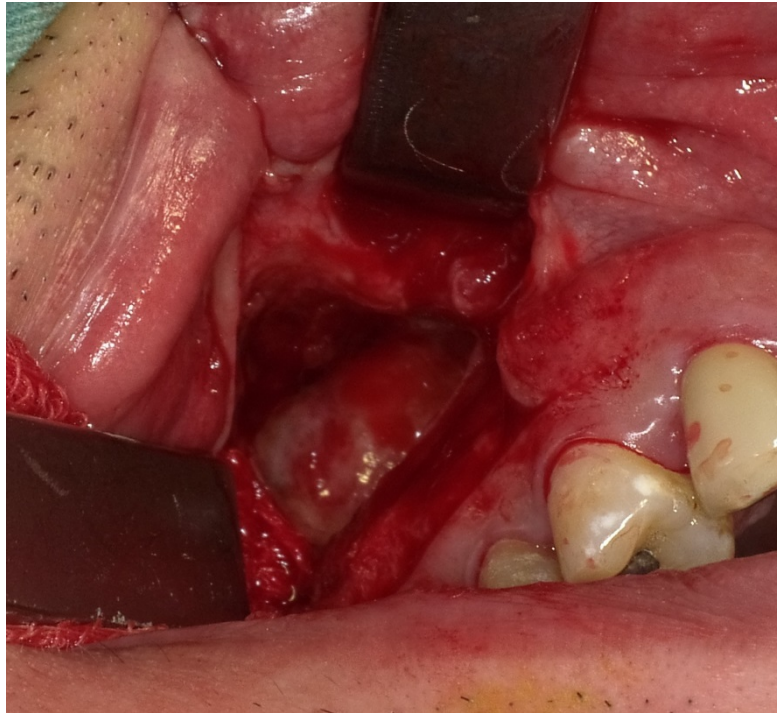


Fig. 2 Intra operative photo showing the cyst in-situ.

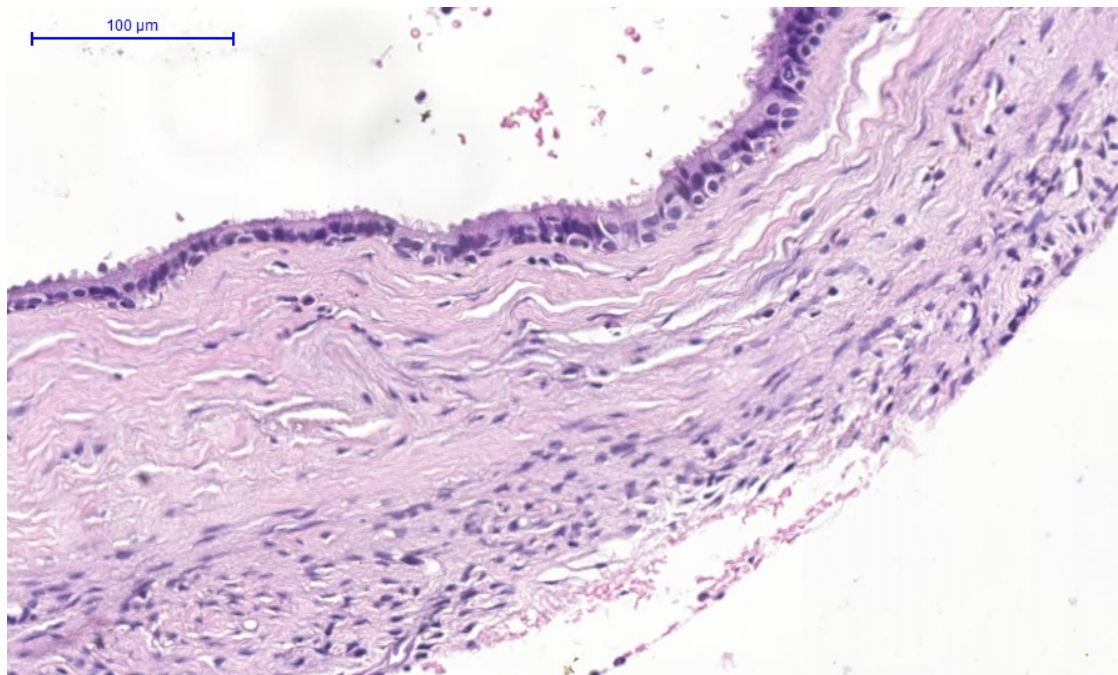


Fig. 3 Photomicrograph of the cyst showing pseudostratified ciliated columnar epithelium lining (original magnification x20, H&E staining).

Discussion

Surgical ciliated cyst of the maxilla was first reported by Kubo in 1927 as a cyst arising from surgery for chronic maxillary sinusitis (Kubo, 1927). Since then, more cases were reported not only as a result of surgery for chronic maxillary sinusitis such as Caldwell-Luc antrostomy but also LeFort I osteotomy in orthognathic surgery (Amin *et al.*, 2003), traumatic tooth extraction, midface fracture (Bulut *et al.*, 2010, An and Zhang, 2014) and alveolar bone grafting (Lekkas *et al.*, 2001). Similar surgical ciliated cysts were also reported in the mandible mainly as a result of orthognathic surgery and genioplasty (Ragsdale *et al.*, 2009).

Pathogenesis of the cyst development was thought to be due to entrapment of remnants of sinus mucosa in the wound after maxillary sinus surgery, or early closure of the natural ostium before the sinus is completely filled with regenerating granulation tissue (Kaneshiro *et al.*, 1981, Maruyama *et al.*, 2002). Small segment of maxillary sinus mucosa may be trapped and resulted in formation of cystic lesion many years later. Following the entrapment, inflammatory process induces cystic changes within the trapped respiratory mucosa leading to cyst formation. The cyst then expands as a result of osmotic differences from surrounding tissue (Kaneshiro *et al.*, 1981, Maruyama *et al.*, 2002).

Clinically, this cyst is a locally aggressive lesion. It can develop many years after the initial surgery to the maxillary sinus. This period ranged from 4 years to 49 years (Bulut *et al.*, 2010). In its initial stage, it's asymptomatic but can progressively cause pain as it expands (Bulut *et al.*, 2010). Pain is aggravated if the cyst is infected. In such cases, at times there may be discharge and fistula formation. It can eventually cause an expansile swelling of the maxillary alveolus and palate (Amin *et al.*, 2003). The expansion is usually towards anterolateral wall of the canine fossa but it may also extend toward the nasal wall or sphenopalatine wall of the sinus (Cano *et*

al., 2009). Teeth involved by the cyst may become non-vital as the expanding cyst at the apical region may affect the neurovascular supply (Leung *et al.*, 2012). In the present case, the patient presented with all the above mentioned typical features except for fistula and loss of vitality of involved teeth.

Radiographically, it is a well-defined unilocular radiolucency closely associated with then maxillary sinus (Amin *et al.*, 2003). The cyst may be surrounded by a zone of sclerosis. As the cyst enlarges, adjacent sinus wall may become thin and eventually perforated (Bulut *et al.*, 2010). Although it is usually unilocular, multilocular variants have also been reported (Pe *et al.*, 1990). Under the microscope, the cyst is lined by pseudostratified ciliated columnar epithelium of respiratory type (Amin *et al.*, 2003). Transition to simple columnar, cuboidal or even squamous epithelium has been observed. Maruyama *et al.* (2002) studied 360 epithelial lining of postoperative maxillary cysts and found that 66% of their length was pseudostratified ciliated epithelium, 28% transition epithelium and 6% squamous epithelium. Goblet cells were present except in areas of squamous epithelium (Maruyama *et al.*, 2002). They also found that the number of goblet cells correlated with inflammatory cells and sialylated glycoconjugates derived from lecithins in goblet cells correlated with cyst wall inflammation and cyst growth (Maruyama *et al.*, 2002). Squamous metaplasia may occur if the cyst was infected (Leung *et al.*, 2012). Cyst contents are mainly hyaluronic acid and heparin sulphate with lesser amount of chondroitin-4-sulphate (Amin *et al.*, 2003). In the present case, the typical radiographic features were observed. However, histologically only the hallmark pseudostratified ciliated columnar epithelium with mucous cell metaplasia was seen in the present case. Squamous metaplasia was not observed in this case since there was no history of infection of the cyst.

In view of its benign nature, surgical ciliated cyst is adequately treated with

enucleation of the cyst. This has remained the treatment of choice whenever clinically possible. In cases of large unilocular cyst with extensive bony perforation, marsupialisation may be performed.

Although surgical ciliated cyst of the maxilla is rarely encountered, surgeons should be aware of such possible complication and able to diagnose at earlier stage of the lesion owing to its locally aggressive nature.

The incidence of surgical ciliated cyst of the maxilla secondary to maxillary sinus surgeries should be declining as a result of conservative management of sinusitis and the advent of endoscopic techniques to treat antral disease (Psaltis *et al.*, 2012). In the contrarily, the incidence due to orthognathic surgery may increase as a result of increasing demand for the jaw corrective surgeries. Although prevention of implantation of sinus mucosa into mandible during a bimaxillary surgery can be achieved by using a different saw blade, prevention of sinus mucosa entrapment during a LeFort I osteotomy is difficult.

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