

Case Report

Maxillary Odontogenic Keratocyst Superimposed by Infection: Case Report

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Introduction

Odontogenic Keratocysts (OKCs) are rare cystic lesions known for their aggressive behavior and tendency to recur after excision. These benign tumors usually occur in the mandible or maxilla and are believed to arise from the dental lamina. The medical community is still debating whether these lesions are developmental or neoplastic [1]. However, most experts agree that they are locally aggressive and require prompt treatment to avoid complications. On imaging, they typically appear as a solitary unilocular lesion that extends longitudinally in the posterior portions of the mandible. Although most are solitary, multiple odontogenic keratocysts can also be present in some cases, and an associated condition such as basal cell nevus syndrome should be considered. It's worth noting that involvement of the maxillary sinus by OKC is rare, but it can happen [2]. The maxillary sinus is part of the paranasal sinuses, which are in proximity to developing tooth and root apices of premolar and molars. The overlapping of various structures makes it challenging to diagnose such lesions via maxillary radiographs. Therefore, prompt diagnosis and treatment of OKCs is essential to prevent further complications [3].

Case presentation

A 22-year-old male patient visited Khartoum Dental Teaching Hospital with complaints of headache, toothache in the posterior left maxilla that radiated to the ear, sinusitis symptoms, and

nasal discharge. Clinical examination revealed a draining sinus on the left maxillary buccal vestibule, from which pus was oozing out, indicating an infection. Straw yellow fluid, as well as cheesy and thick contents, were noticed in the pus. A radiographic examination was done using CBCT (Cone Beam Computed Tomography), which showed a well-circumscribed lytic lesion in the posterior left maxilla extending from the maxillary tuberosity to the level of the first premolar and involving the maxillary sinus. An impacted wisdom tooth was also noticed above the premolars roots with its crown downward and distally. Subsequently, incision and drainage were performed, followed by antibiotic coverage and daily dressing as irrigation with warm saline until the pus disappeared. The patient was then scheduled for an excisional biopsy of the cystic lining. A sulcular incision with mesial releasing incision was made, and the buccal wall was found to be already perforated. The cystic lining was enucleated with periosteotomy, and the associated impacted left maxillary wisdom tooth was also removed. The roots of the associated teeth were also curetted, and the cavity was irrigated with saline. The flap was sutured with 4.0 vicryl suture, and a pressure pack was applied to the corresponding cheek. The sinus membrane was left in its place, and the patient was prescribed antibiotics, anti-inflammatory, and decongestant drugs. The patient returned for follow-up after 7 days with no symptoms of sinusitis or any other complaints.

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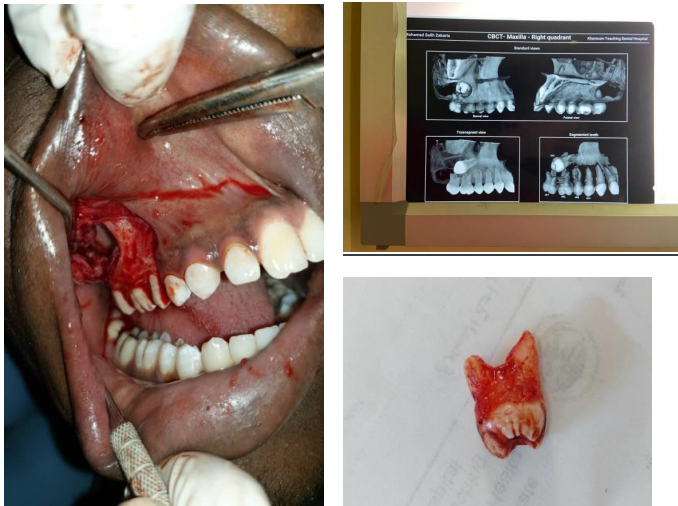


Figure 1: Showing intra-operative photo, CBCT and impacted wisdom tooth.

Histopathology

Our lab received fragmented cystic lining with areas of variable thickness measuring in aggregate 6x6x2 cm. Representative sample was submitted in three blocks.

Microscopy of the adequate sample of soft tissue submitted showed in low power a cystic lining of variable thickness lined by parakeratinized stratified squamous epithelium with a corrugated surface tending to separate from the underlying connective tissue capsule. The high power view featured an Odontogenic cyst lined by 7-11 layers of parakeratinized Odontogenic epithelium with a well palasaded hyperchromatic basal layer the epithelial lining was devoid of rete ridges. The underlying connective tissue capsule exhibited multiple daughter cyst scattered in the area. According to the above mentioned findings a diagnosis of Odontogenic keratocyst was rendered.

Discussion

It seems like odontogenic keratocysts are commonly found in younger patients, usually in their second or third decade of life [1,2]. These cysts can appear in either the mandible or maxilla, with a male bias. Although odontogenic keratocysts are more frequently found in the mandible, it is rare to find them in the maxilla. As the cyst grows, it can encroach on the space of the sinus and displace its borders, which could lead to the "ectopic" eruption of a tooth [3]. Radiographically, odontogenic keratocysts typically show as well-defined unilocular or multilocular radiolucencies with corticated margins. While unilocular lesions are more common, the multilocular variant is seen in about 30% of cases, mostly in the mandibular arch [4,5]. Odontogenic keratocysts are developmental cysts that arise from cell rests of the dental lamina, which is the oral epithelial lining of the developing tooth follicle [6]. Some experts consider them to be a benign cystic neoplasm rather than a developmental cyst [7]. Although these cysts are most common in males during their second and third decade of life, some studies have reported a bimodal age distribution with an additional peak in the fifth

and sixth decades [8]. Maxillary sinus odontogenic keratocyst is rare, comprising less than 1% of all reported cases, and has unique features compared to other cysts of the maxillofacial region. This case report describes an unusual case of invasive maxillary sinus odontogenic keratocyst that extended into the orbital floor, pterygoid plates, and hard palate in a 30-year-old female. The report emphasizes the need for extensive treatment of cystic maxillary sinus lesions, regardless of the nature of the lesion, due to the site's high susceptibility to secondary infection and recurrence. Additionally, the case establishes a set of imaging modalities and specific treatment approaches to be followed for maxillary sinus odontogenic keratocyst based on the literature of all the previous reported cases [9].

Conclusion

Despite that Maxillary OKC is of rare occurrence, OKC should be considered frequently in the differential diagnosis of maxillary cystic lesions. Displacement of impacted tooth by keratocyte can change the position of the tooth into the maxillary sinus with development of signs of sinusitis and increase the chance of infection.

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