CASE REPORT

Ciliated cyst of the maxilla following trauma: An unusual case report

Harsha Vardhan Gowthammnath, J. S. Jesija, K. Saraswathi Gopal
Oral Medicine and Radiology, Meenakshi Ammal Dental College, Chennai, Tamil Nadu, India

Abstract
Surgical ciliated cysts or post-operative maxillary cysts are benign cystic lesions usually induced after a surgical procedure in the maxillofacial area. It is a cystic lesion that develops, following a radical sinus surgery to treat maxillary sinusitis. The cyst progressively enlarges due to the osmotic difference causing destruction of the adjacent bone and structures. The clinical scenario may be identical to a radicular cyst if a nonvital tooth is involved. This is a case report of a healthy 17-year-old adolescent with the complaint of swelling in the right side of the face for the past 2 years.

Keywords: Ciliated cyst, maxilla, radiolucent, trauma

Introduction
The surgical ciliated cyst has been well-known since 1927, and it is documented in the Japanese literature.[1] The surgical ciliated cyst is also known as post-operative maxillary cyst, post-operative paranasal cyst, and surgical ciliated cyst of the maxilla.[2,3] Most of these cysts have occurred as delayed complication of radical maxillary sinus surgeries to treat various sinus pathologies.[3,4] This is a case report of a ciliated cyst of the maxilla that had developed in a young adolescent boy following minor traumatic injury.

Case Report
A 17-year-old male presented to the Department of Oral Medicine and Radiology with the complaint of swelling in the right side of the face for the past 2 years. The patient presented with a history of trauma 2 years ago, when he was hit by a rod while travelling in a bus. Following the injury, he developed a swelling in the right side of his face. There were no secondary changes during this 2-year period. The patient had developed pain in the upper right anterior teeth region 2 months ago. He had consulted a private dentist, following which a root canal therapy was initiated, but left incomplete, with no pain thereafter.

On clinical examination, the patient presented with a diffuse swelling in the right side mid face region approximately measuring 3 cm × 3 cm, extending superiorly one cm below the infra-orbital margins and inferiorly 0.5 cm above an imaginary line joining the corner of right lip to the ear lobe. Anteriorly, the swelling was found to diffuse with the ala of the nose and posteriorly it extended 3 cm short of the ear lobe. The swelling was hard and nontender to palpation [Figure 1a]. Intra-orally, there was obliteration of the buccal vestibule in relation to 12 and 13 with no secondary changes [Figure 1b]. Vitality test was performed from right first premolar to left canine and 11, 12, and 13 showed no response to the test and proved to be nonvital. Based on the history and clinical examination, a clinical diagnosis of benign odontogenic cyst or tumor was considered. The patient was then subjected to radiological evaluation.

Orthopantamogram revealed a single large unilocular radiolucency measuring approximately 3 cms in diameter in the right maxillary region extending superiorly to the level of maxillary antrum and inferiorly toward the alveolar process in relation to 12, 13, 14, 15, and 16 which obscured the right maxillary sinus. There was also evidence of apical displacement of the tooth 13 [Figure 2].

Computed tomography (CT) maxilla revealed evidence of a well-defined, moderate-sized expansive cystic lesion with thin bony rim and hypo dense soft tissue component arising from the alveolar margins of right maxilla projecting into the adjacent right maxillary sinus almost completely occluding its lumen, predominantly expanding in the labial aspect, suggestive of odontogenic cyst [Figure 3].

Following the above mentioned investigations, fine-needle aspiration cytology was performed by creating a surgical
window. It yielded a straw colored aspirate which showed numerous polygonally shaped, anucleated and nucleated epithelial cells along with inflammatory cells, predominantly lymphocytes, few eosinophils, plasma cells, and macrophages in a background of red blood cells suggestive of infected cystic material.

Subsequently, incisional biopsy was performed, and histopathology revealed cystic lining epithelium of varying thickness supported by fibro vascular connective tissue showing chronic inflammatory cell infiltrate predominantly of lymphocytes and plasma cells. The cystic lining epithelium was not continuous and was of stratified squamous type with areas exhibiting flattened epithelial cells to cuboidal cells intermixed with vacuolated cells, suggestive of the infected cyst.

With the results of both aspiration and incisional biopsy being suggestive of a cystic lesion, complete surgical excision of the cyst was planned under general anesthesia. The excised specimen in toto was submitted for histopathological evaluation. The microscopic features revealed pseudostratiﬁed ciliated columnar epithelium predominantly, and few areas exhibiting 2-4 cell layered thick stratified cuboidal/squamous nonkeratinized epithelium associated with fibrovascular connective tissue. The connective tissue exhibited areas of inﬂammation, seromucinous acini, extensive ﬁbrosis, areas showing irregular bony trabeculae with osteocytes, and extensive hemorrhage suggestive of ciliated cyst of the maxilla [Figure 4].

**Discussion**

Radiolucent lesions involving the jaws are of utmost concern to the clinician. These lesions range from developmental disturbances to reactive or inﬂammatory processes to lesions of benign or malignant origin. Irrespective of the etiology of radiolucent lesions, they must be investigated and actively pursued in order to treat them appropriately.

In the present case report, the radiolucent lesion represented a ciliated cyst of the maxilla. Clinically, it is commonly seen among middle-aged patients.[1] The presenting complaints are usually swelling pain or discomfort involving the maxilla. The swelling may either present extra-orally or intra-orally with radiographs revealing a well-deﬁned radiolucent area anatomically distinct from the maxillary sinus.[1]

A detailed review of the literature with very few exceptions suggested that ciliated cysts are usually post-operative. Gregory and Shafer (1958) reported cysts following operations for maxillary sinusitis, particularly the Cald-well Luc approach including nasal antrostomy. Wassmund (1939) has published two case reports on similar cysts arising after gunshot injuries.[2] Shuttleworth and King (1951) have reported these cysts following malar slash maxillary fractures.[5] With regard to the pathogenesis, Kubo (1927 and 1933) postulated that the lesion originated from the trapped sinus mucosa in the wound either from a surgical procedure or as a result of trauma.[6] Imai (1933) hypothesized that mucosa derived from the nasal cavity regenerated within the antrum.[7] Further, Mohri et al. (1977) attributed the origin of the lesions to the closure of both the natural ostium and the intranasal opening following a surgical procedure.[8]
The conventionally termed “surgical ciliated cyst” is commonly encountered among the Japanese population but appears to be a rare entity in the most other parts of the world.[9] It has a slight male predilection with a wide presenting age range from 21 to 80 years, and it is thought to occur 3-61 years following a maxillary surgical procedure.[10]

Radiographs reveal well-defined radiolucent areas in close relation to the maxillary sinus. In a series of cases reported by Yamamoto, Takagi most of the cases were unicocular with very few showing a multilocular presentation.[4] The buccal cortical bone is usually intact with surrounding bone sclerosis evident among few cases. Kaneshiro et al. reported missing margins in the lateral and posterior walls of the lesion. Occasionally, the cystic area encroaches into the sinus but it lacks the communication between the two which has been demonstrated by injecting a radiopaque material (Shafer et al. 1983). In the early diseased stage, there is no bony destruction, but as they expand the sinus wall, it becomes thinned out and eventually gets perforated mimicking a malignant neoplasm.[9,11]

Histologically, these cysts are usually lined by pseudostratified ciliated columnar epithelium, with squamous metaplasia within the chronically inflamed areas. A combination of ciliated, cuboidal, and squamous epithelium with varying numbers and layers of mucous cells may be seen. The underlying connective tissue can be cellular or fibrotic.[11] Extensive hemorrhage and foci of calcification may also be present. Mucous retention cyst of the antrum should be considered as one of the differential diagnoses as they are also lined by pseudostratified columnar epithelium. However, they do not follow an aggressive course, nor do they tend to present clinically as a swelling. They are usually diagnosed as incidental findings in an asymptomatic patient. Gardner (1984) characterizes these harmless lesions as pseudocysts.”[12]

To conclude, the most widely accepted theory of pathogenesis is thought to be due to the entrapment of the sinus mucosa following surgery (traumatic theory).[13] Our case stands unique with a history of trauma 2 years ago resulting in an identical pathology.

References