Persistent Deciduous Molar as an Etiology for a Maxillary Sinus Mucocele

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Abstract Mucoceles are accumulations of trapped mucus, forming cystic expansile lesions. Maxillary sinus mucoceles are rare amongst paranasal sinus mucoceles, usually being a late sequel of Caldwell-Luc surgery. We present a case of a maxillary sinus mucocele due to a persistent carious deciduous molar in a middle aged patient, reported to highlight its unusual etiology.

Keywords Mucocele · Persistent deciduous molar

Introduction

Mucoceleles are chronic, benign, mucus-filled cystic lesions in the paranasal sinuses caused by obstruction of sinus ostia. Expansion from pressure and inflammation leads to remodeling and erosion of bone, changing the bony architecture [1]. The etiology is varied; the case presented here is a sequel of an infected deciduous tooth persisting into adulthood.

Case Report

A 48-year-old male patient reported in the ENT OPD, with a history of slow growing, initially painful swelling of right cheek for 5 months and right sided nasal obstruction for 3 months. It was preceded by severe toothache of the right upper first premolar and fever for 15 days. He was treated at a peripheral health center with aspiration, antibiotics and

anti-inflammatory drugs, following which pain subsided, but the swelling persisted.

The swelling was diffuse, about 7.5 cm × 5 cm in size, firm, non tender, extending deep to the right ala and nasal cavity causing a smooth bulge. It also extended into the right upper gingivo-buccal and gingivo-labial sulcus, from the canine to the first molar tooth, with a small area of ulceration over the site of aspiration. The upper right premolars were malaligned, carious and eroded (Fig. 1). Nasal endoscope couldn't be passed on the affected side.

Axial and coronal CT (Fig. 2) showed an expansile cystic lesion involving the entire right maxillary sinus, its medial wall touching the nasal septum, posterior end of the swelling extending into the nasopharynx up to the pterygoid plate. The entire sinus was filled with a fluid shadow without area of air shadow and scattered areas of demineralization of the sinus walls.

FNA yielded hemorrhagic transparent fluid. Cytological and microbiological examination of the fluid revealed presence of inflammatory cells without cellular atypia or microorganisms.

The swelling was provisionally diagnosed as a dental cyst/mucocele of maxillary sinus. Under general anesthesia, a sub-labial Caldwell-Luc's antrostomy was done and the antral cavity was cleared of the fluid filled sac. Most of the sinus walls were thick, fibrotic and expansile making it floppy and redundant after drainage of the tense collection. The lining of the sinus was smooth and featureless except around the floor where a carious persistent deciduous right first molar (evidenced by the presence of three roots and a buccal protrusion of enamel bulge) was protruding into the sinus cavity. It was extracted and bone saucerized. The right upper 2nd permanent premolar was carious and extracted resulting in an oro-antral fistula, which was repaired with a palatal flap. Redundant tissue on the medial

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Fig. 1 Carious and malaligned upper right premolars with offending deciduous first molar (*circled*)

Fig. 2 Axial and coronal CT showing the expansile cystic lesion involving the right maxillary sinus, causing nasal obstruction, extending into the nasopharynx up to the pterygoid plate. There is no air shadow. Scattered areas of demineralization of the sinus walls are seen

wall was quilted. There were no ostia visible, so an inferior meatal antrostomy was done.

Histopathology of the maxillary sinus walls showed an exudate of neutrophils and macrophages, enmeshed in fibrin with foci of hemorrhage. No lining epithelium or granulation tissue was seen. HE stain did not reveal any organism. The histological impression was thus of an abscess, possibly infected mucocoele.

Discussion

A mucocoele is a cyst developing from a sinus mucosa most commonly the frontal or ethmoidal sinuses. Maxillary sinus

mucocoeles are exceptional, having been reported in only 3–10% of cases [2]. Mucocoeles are consequent to an obstruction of the sinus ostia and drainage pattern, with accumulation of mucus within the sinus cavity. Continual accumulation causes it to expand from the pressure. Although, mucocoeles are usually sterile, pain may indicate infection [1, 3]. In the reported case the contents were sterile and in our opinion it began as a pyocoele secondary to the dental infection as evidenced by the initial presence of pain in tooth as well as the swelling and then after conservative treatment, it turned into a mucocoele. The pus caused a desmoplastic reaction in the walls of the sinus and so the walls of the mucocoele in the patient were thick unlike the usually thinner walls of a mucocoele.

Sinus walls may be remodeled or completely de-ossified and eroded. The factors involved in this process include cytokines released from lymphocytes and monocytes, due to sinus obstruction and superimposed infection. The cytokine release stimulates fibroblasts to secrete prostaglandins and collagenases, which in turn stimulate bone resorption leading to expansion of the mucocoele [4]. In our case, the thick, fibrous and floppy medial wall of the antrum had to be excised and plicated. Histologically, mucocoeles may be primary or secondary. Primary ones are in fact mucus retention cysts while secondary ones are “true” mucocoeles. They are lined by pseudostratified columnar epithelium, occurring when the sinus ostium is obstructed [5]. Our patient had a “true” mucocoele secondary to the obstruction of the natural ostium of the maxillary sinus which had occurred due to the desmoplastic reaction on the sinus walls following a suppurative inflammation.

The various known etiologies of maxillary sinus mucocoeles, include chronic infection, allergic sinonasal disease, trauma, previous surgery. However, in some cases cause remains uncertain [5, 6]. Busaba and Salman compared the bacteriology of maxillary sinus mucocoeles to chronic sinusitis and reported that the data do not support infection as the main origin of nontraumatic maxillary sinus mucocoele [6]. A Turkish report studying the clinical presentation of maxillary sinus mucocoeles in 14 cases found that 36% patients had a history of previous surgery leading to maxillary mucocoele formation, while in 64% cases no known pathology could be discerned [1]. A Japanese study found that the possible causes leading to simulating cases can be; unknown (39.3%), recurrent infection in the postoperative cavity (28.6%), dental origin (10.7%), atypical neuralgia (10.7%) and shortly after a preceding sinus surgery (within 12 months, 10.7%) [7]. In our case, infection originating from a persistent carious deciduous molar tooth was the apparent etiology.

CT is the preferred imaging modality where the mucocoeles appears as an expanded, airless sinus filled with

homogeneous material. The walls of the sinus may be either normal or remodeled, with thickening, thinning and erosion to various degrees, often within the same sinus. The distinction between a mucocoele and a mucous retention cyst can be made by the presence of air outlining the upper surface of the retention cyst [8]. Mucocoeles may be treated by either radical surgery or conservative surgery. Radical surgery entails the complete extirpation of the mucus membrane with obliteration or cranialization of the sinus cavity. Conservative surgery involves marsupialization of the mucocoele with maintenance of adequate sinus drainage, to minimize risk of recurrence. This approach is based on the principle that the underlying disturbance in a mucocoele is one of blocked ostial drainage and not of a diseased mucous membrane and is now preferably done endoscopically. Marsupialization of the mucocoele with establishment of ostial drainage relieves the symptoms of the mucocoele and prevents re-accumulation of the mucus later [9].

We had, in our patient preferred marsupialization with an inferior meatal antrostomy. A middle meatal antrostomy was not preferred because the medial wall of the maxillary sinus was thick and featureless making it difficult to locate the site of the natural ostium. Moreover as the adequacy of the mucociliary clearance mechanism was doubtful we made an inferior meatal antrostomy for gravitational drainage. Once adequate drainage is established, the recurrences are rare, which occur many years after the surgery.

Conclusion

Maxillary sinus mucocoeles are the rarest amongst paranasal sinus mucocoeles. Their etiology is varied, the commonest

being post-surgical. Dental causes are unusual. A misaligned deciduous carious tooth can be an important precipitating cause of such a condition. So a persisting deciduous tooth if carious should be extracted as conservative treatment in such cases can act as a precursor for a maxillary sinus mucocoele.

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