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Case Report:

A Rare Presentation of Odontogenic Keratocyst Mimicking an Antral Polyp.

Authors
S Sudhakar, Reader,
V Geethika,Post graduate Student,
P Ramaswamy, Professor & HOD,
B Praveen Kumar, Reader,
P Sreenivasulu, Senior Lecturer
Department of Oral medicine & Radiology, St.Joseph dental college & Hospital, Eluru, Andhrapradesh, India.

Address for Correspondence
Dr. Sudhakar S,
Reader, Department of Oral medicine & Radiology,
St.Joseph dental college & Hospital,
Eluru, Andhrapradesh, India.
E-mail: drsudhakaroralmed@yahoo.co.in

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Abstract: Maxillary sinus harbours many pathological lesions. Many of those presents as a sinonasal mass and are rarely symptomatic. These masses are usually an antral polyp, mucoceles or mucous retention cysts. Odontogenic keratocyst, a benign odontogenic lesion presenting within the maxillary sinus is a rare entity. We present a case of odontogenic keratocyst of the maxillary sinus in a 35 years old female.

Key Words: Keratocyst; Maxillary sinus

Case Report:

A 35 years old female patient reported with a complaint of discomfort on the upper right cheek region since 15 days. Patient history suggested that she had similar complaints for the past three months and had subsided upon taking some over the counter medications. The patient claims the present complaint is since 15 days and had not subsided upon taking the same medications. Although the discomfort was not debilitating nor it is causing any functional disability she reported to us in search of a permanent relief. Her medical, surgical and family histories were non-contributory. The past dental history revealed history of extraction of upper right second premolar in a private dental clinic a year ago, following dental caries and periapical infection. The post-operative period was uneventful.

On extraoral examination mild tenderness over the right maxillary sinus region was elicited. Nares and nasal septum appeared normal. Intraoral examination showed missing right second premolar and a scar/ indentation noticed over the premolars vestibular space with mild tenderness on palpation. There was no evidence of vestibular obliteration, expansion or pus discharge (Fig 1).

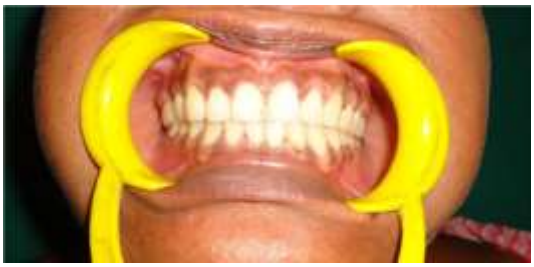

Fig 1: Intraoral photograph showing the normal vestibular spaces

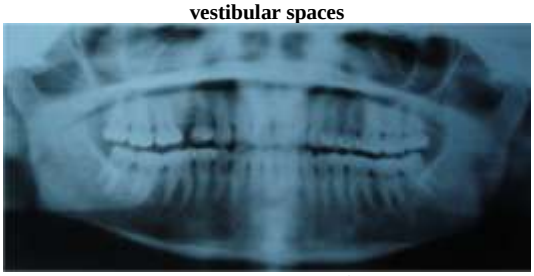

Fig 2: OPG showing the healing defect over the right premolar region

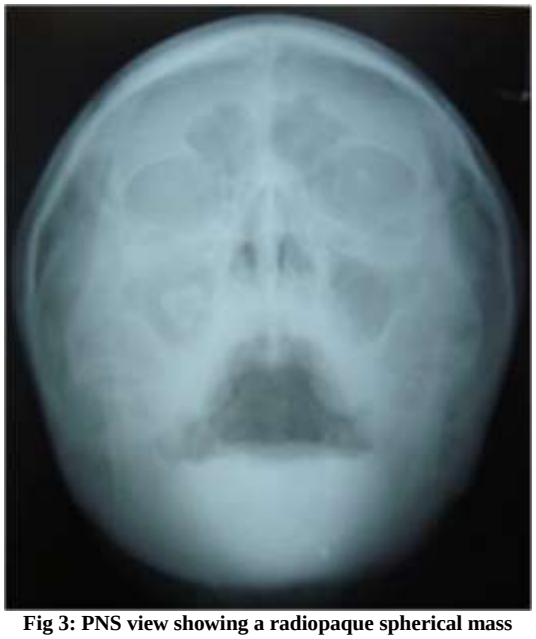

Fig 3: PNS view showing a radiopaque spherical mass involving the right maxillary sinus

Fig 4: Computed tomography picture showing a well defined spherical area involving the right maxillary sinus

Fig 5: Intra-operative photograph

Patient was then subjected to routine orthopantomograph (OPG) and paranasal sinus (PNS) view. The OPG showed missing right second premolar with an arc shaped bone loss over the right first premolar extending from the level of alveolar crest towards the junction of cervical and middle third of the root (Fig 2). The PNS view showed a well-defined pyramidal shaped radiopaque mass ranging 1.5x1 cm in size located at the core of the right maxillary antrum (Fig 3). The computed tomography picture showed presence of a well defined homogenous spherical mass ranging 2x2 cm within the confines of the right maxillary sinus. The surrounding area appeared normal with no evidence of bony destruction (Fig 4). The features were suggestive of a polypoid mucosal mass within the right antrum.

Fig 6: Photomicrograph of the excised specimen

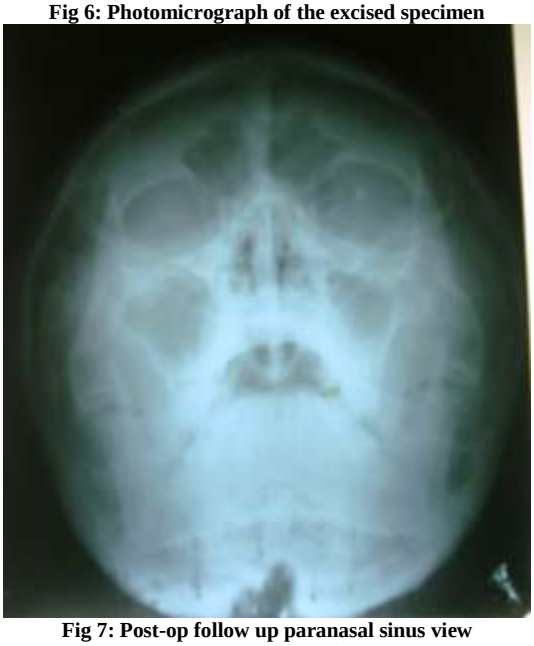

Fig 7: Post-op follow up paranasal sinus view

Considering the symptomatic nature of the mass, the panel of experts comprising oral physician, oral surgeon and otorhinolaryngologist decided to surgically explore the lesion. Under general anesthesia, the mass was approached through maxillary antrostomy and completely enucleated (Fig 5). The post-operative period was uneventful.

Histopathological analysis of the excised mass showed presence of pseudostratified ciliated columnar epithelium with subepithelial glands, numerous capillaries, inflammatory infiltrates and bony trabeculae. Focal areas showed presence of stratified parakeratinized corrugated epithelium of 5-6 layers thickness with palisading of basal tall columnar cells. The connective tissue shows mild chronic inflammatory cells (Fig 6). These features were suggestive of odontogenic keratocyst- parakeratinized variant.

Considering the above diagnosis the patient was recalled and cautioned about the recurrent nature of the lesion. Till date there is report of recurrence and the patient is kept under every six months follow-up (Fig 7).

Discussion:

The diseases involving the sino-nasal regions are manifold and it includes developmental, infectious, traumatic and even malignancies. Interestingly the clinical presentations too

range from completely asymptomatic to symptoms like pain, swelling, obstructions, discharge and altered smell etc., The commonest sino-nasal disorders are sinusitis, allergic rhinitis and antrochoanal polyps. The sinusitis and rhinitis are inflammatory lesions of their respective regions whereas the antrachonal polyp is considered to be its sequel.

The common differential diagnosis of ACPs should include juvenile angiofibroma, nasal glioma, meningoencephalocele, inverted papilloma, mucocele, mucus retention cyst, Tornwalt's cyst, grossly enlarged adenoids, lymphoma and nasopharyngeal malignancies.(1-6) Presence of odontogenic keratocyst (OKC), a lesion arising from the odontogenic apparatus, a completely non-native entity in the sinonasal region is a rarity. Until now, only two cases of OKC present completely within the maxillary sinus has been reported in the literature.(7)

OKC, first described by Philipsen in 1956 and later designed by the World Health Organization as keratocystic odontogenic tumor (KCOT), is a benign uni or multicystic intraosseous tumor of odontogenic origin, with a characteristic lining of parakeratinized squamous epithelium.(8)

The origin of OKC is suggested to be from the dental lamina remains.(9) However, some author's support that it should be considered a benign cystic neoplasm related to the mutation of tumor suppressor gene, PTCH and more recently, intracystic fluid pressure was found to be involved in OKC growth.(10,11)

OKC comprises approximately 11% of all cysts of the jaws. The OKC has a predilection for men and are common in second and third decades of life. They occur most commonly in the mandible, especially in the posterior body and ramus regions.(12,13) They almost always occur within the bone, although a small number of cases of peripheral variants have also been reported.(8)

Reports suggests that less than 1% of all cases of OKC occur in the maxilla with the involvement of the maxillary sinus.(13,14) In the above said cases, the OKC's are found to be primarily developing from the maxillary bone then extends to involve the maxillary sinus. However, rare presentations of OKC completely restricted within the maxillary sinus without the involvement of the alveolar bone or in association with an unerupted teeth have also been reported.(7)

The case reported here also showed OKC present within the sinus but more interestingly without the association of any unerupted/ impacted tooth. Usually patients with OKC presents with swelling, pain and discharge. Distinctive clinical features include a potential for expansion, local destruction and a tendency for multiplicity, our patient did not have any such presentation.(15)

Radiographically, intraosseous odontogenic keratocyst present as a multilocular or unilocular radiolucency with well-developed sclerotic borders. As OKC appearance within the maxillary sinus is rare, its radiographic image in such situation may be misinterpreted, as seen with the present case. Although computed tomography can provide information on the extent of these lesions, due to relative lack of specific characteristics it may not contribute to diagnosis and preoperative preparation.(9) Contrast enhanced MR imaging can provide the essential macroscopic detail, including focal wall enhancement and iso-intense intraluminal soft tissue mass, which correlates with the histological findings of focal inflammatory ulceration of the cyst lining, cytokeratin and cell debris.(16) In many instances only CT is advised, only rarely both CT and MRI are advised that too when there is a gross destruction of bone, since the distinction between them is more of an intellectual interest rather than clinical significance. Unfortunately in the present case too only CT was advised. Histologically, OKCs are formed with a stratified squamous epithelium that produces orthokeratin, parakeratin or both. The epithelial lining appears corrugated, with a wellpolarized hyperchromatic basal layer and the cells remain basaloid almost to the surface. No rete ridges are present; therefore, the epithelium often sloughs from the connective tissue.

The epithelium is thin and mitotic activity is frequent; therefore, OKCs grow in a neoplastic fashion and not in response to internal pressure. The lumen is frequently filled with a foul smelling cheese-like material that is not pus but rather collected degenerating keratin.(8) Various treatment alternatives based on surgical approaches have been suggested, such as marsupialization, enucleation, enucleation with Carnoy's solution, enucleation with cryotherapy, curettage and resection. Simple enucleation was associated to a higher recurrence rate, while resection and enucleation with bone curettage presented lower rates. Special attention should be given to the dentate area if the enucleation is chosen as treatment, due to higher rates of recurrence found in OKC associated with teeth.(7) In the present case enucleation was done based on the initial judgement of the lesion, however the patient is under regular follow up with no evidence of any recurrence till date.

Conclusion

Odontogenic keratocyst is an uncommon entity in the maxillary sinus region. If involved it usually extends from the underlying maxillary bone then secondarily into the sinonasal region. The case presented here is a rarity as neither there is a gross destruction of the underlying bone nor any associated impacted tooth and the lesion is completed within the sinus. The histopathological analysis suggested the presence of squamous epithelium and no evidence any sinonasal comorbidity.

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