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CASE REPORT

Dentigerous Cyst Associated with an Ectopic Third Molar in the Maxillary Sinus: Report of Cases and Review of Literature

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ABSTRACT

Introduction: The ectopic eruption into the nondentate region such maxillary sinus is rare and scantly documented. In the present study, we report two cases of ectopic teeth in maxillary sinus associated to dentigerous cyst (DC) with their clinical features and surgical management.

Case reports

Case 1: A 30-year-old male was referred with a chief complaint of pain and swelling over right cheek region since 16 months and discomfort of nasal breathing.
Case 2: A 28-year-old female reported to the Department of Oral and Maxillofacial Surgery with a chief complaint of facial asymmetry due to swelling on the right side of the face since 6 months.

Management: Complete enucleation of the cysts along with the impacted third molar was done under general anesthesia via an intraoral approach by exposing the anterolateral wall of maxillary sinus. The specimens were sent for histopathologic examination which confirmed DC.

Conclusion: Complete enucleation of the DC and regular postoperative follow-up with radiographic examination is mandatory to rule out any recurrence.

Keywords: Dentigerous cyst, Ectopic tooth, Maxillary sinus.

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INTRODUCTION

The ectopic eruption into the nondentate region such maxillary sinus is rare and scantly documented. In the present study, we report two cases of ectopic teeth in maxillary sinus associated with DC with their clinical features and surgical management.

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CASE REPORTS

Case 1

A 30-year-old male referred to Department of Oral and Maxillofacial surgery with a chief complaint of pain and swelling over right cheek region since 16 months and discomfort of nasal breathing. Clinical examination revealed that the swelling was soft, fluctuant, painful without any hypoesthesia of infraorbital nerve.

It was also extending from the lateral wall of the nose to zygomatic region mediolaterally and from infraorbital region to right upper lip superoinferiorly. There was obliteration of the nasolabial fold.

Intraorally, the swelling was extending from maxillary right central incisor to the molar region with obliteration of the vestibule. The mucosa overlying the swelling appeared to be intact. The upper right third molar was absent. On computed tomography (CT) scan, the axial and coronal views (Fig. 1) revealed an ectopic tooth situated superiorly to maxillary antrum and posteroinferiorly to the floor of orbit with large cystic lesion occupying the entire maxillary sinus and resorption of his right anterolateral wall. Midfacial degloving (Fig. 2) approach allowed a complete enucleation and extraction of the ectopic molar. The postoperative healing was uneventful. Histological examination of the specimen confirmed the diagnosis of a DC. Postoperative follow-up orthopantomographs were taken at regular monthly intervals. Six months postoperatively, the dimensions of the cystic cavity were reduced both clinically and radiographically. The patient is still under routine follow-up and will be closely monitored for long-term outcome.

Case 2

A 28-year-old female reported to the Department of Oral and Maxillofacial Surgery with a chief complaint of facial asymmetry due to swelling on the right side of the face since 6 months. The swelling was initially small, which gradually increased to the present size (Fig. 3). Intraoral examination revealed absence of the upper right third molar and a diffuse hard swelling over the right maxilla.

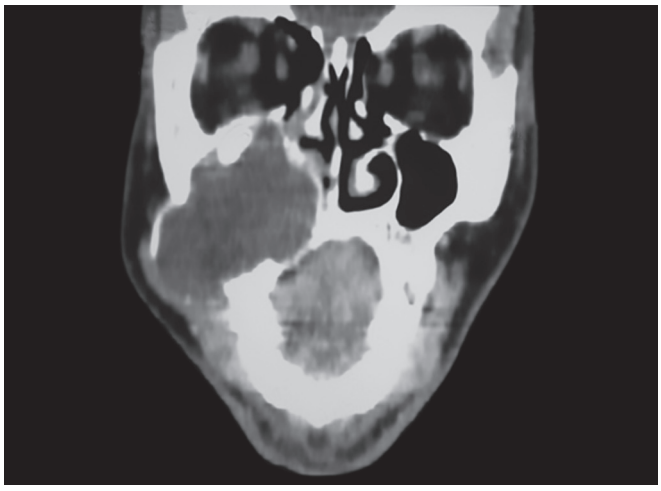

Fig. 1: Coronal CT image showing surrounding a tooth situated superiorly to maxillary antrum and posteroinferiorly to the floor of orbit with an expansile cystic lesion

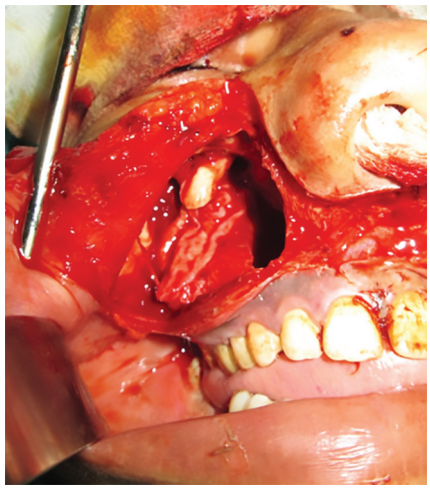

Fig. 2: Surgical view of the cyst enclosing the impacted teeth

Fig. 3: Preoperative clinical photographs showing swellings over right cheek region

Fig. 4: Paranasal sinus view radiograph illustrating an ectopic tooth in the maxillary sinus

Radiograph showed an ectopic tooth along with large cystic lesion occupying the entire maxillary sinus (Figs 4 and 5).

In parallel, physical examination observed that the patient was of short stature and had webbed neck with a low hair line at the back of the neck, and primary amenorrhea (Fig. 6). As Turner syndrome was suggested, an ultrasound of the abdomen was performed, which revealed hypogonadism but no other systemic abnormalities were showed. Finally, chromosomes analysis found a typical karyotype of TS (45XO).

Complete enucleation of the cyst along with the impacted third molar was done under general anesthesia via an intraoral approach by exposing the anterolateral wall of maxillary sinus. The specimen was sent for histopathologic examination which confirmed DC. Postoperative healing was uneventful. Our patient was referred to endocrinology department and was simultaneously followed-up over a period of next 3 years, and there were no signs of recurrence.

Fig. 5: Axial CT SCAN showing maxillary radiolucent lesion surrounding the right impacted third molar

DISCUSSION

Ectopic teeth in nondentate region of the maxillofacial skeleton may be present in the palate, maxillary sinus,

Fig. 6: Clinical picture showing TURNER syndrome phenotype (short stature, webbed neck and undeveloped breasts)

mandibular condyle, coronoid process, orbit, nasal cavity, or even the skin. This phenomenon is rare. On a PubMed search, only about 59 cases of ectopic tooth in the maxillary sinus were published from 1950 to January 2014 in the English literature.[1]

A review by Lamb et al[13] identified only 35 reported cases of this phenomenon in English language medical literature since 1927. Dentigerous cyst associated with ectopic tooth within the maxillary sinus is fairly rare and only 17 cases 8 had been reported since 1980.[2]

The exact etiology of ectopic tooth in the maxillary sinus is not clear.[1] Tooth development involves complex interactions between the oral epithelium and the underlying mesenchymal tissue.[3] If abnormal tissue interactions disrupt the process, the result is ectopic tooth development and eruption. Some reports have suggested relationship with DCs.[2-5]

The displacement of the tooth may be due to the pressure caused by the cystic enlargement. Other etiology may include developmental disorders, such as cleft palate trauma causing displacement of the teeth, maxillary infection, crowding, high bone density, and genetic factors.[6]

Our observation of Turner syndrome is an example of such genetic etiology which had never been described in the literature. Indeed, this genetic disorder is caused by numerical or structural aberration of the X chromosome; sex chromosomes contain genes for tooth development. The most frequent oral findings in patients with Turner syndrome are hypoplastic mandible, high palatal vault, small teeth, early eruption of permanent teeth, and short roots.[7]

The term "dentigerous cyst" was coined by Paget in 1853. These cysts are the most common type of developmental odontogenic cysts arising from the crowns of impacted, embedded, or unerupted teeth.[8]

The most reasonable theory to explain its pathogenesis appears to be that the cyst is the result of the accumulation of fluid between an unerupted tooth and the surrounding reduced enamel epithelium. They are two times common in males than in females. About 70% of DCs occur in the mandible and 30% in the maxilla.[9]

Dentigerous cyst associated with an ectopic tooth within the maxillary sinus is fairly rare, and only 20 cases had been reported in MEDLINE since 1980, including the three cases reported by Buyukkurt et al.[5,10]

According to a recent literature review of reports by Beriat et al,[11] there were 18 male patients and 12 female patients. There is a higher incidence in men than in women, and the condition is usually diagnosed in second to third decade of life.[1]

The most frequent symptoms are facial pain, facial edema rhinorrhea, headache, and nasolacrimal obstruction.[12] A large maxillary cyst involves the whole sinus and can transmit pressure to the walls of sinus; consequently, ophthalmologic and nasal symptoms may develop. Moreover, various findings are described: Epiphora, superior orbital fissure syndrome, eruption into the orbit causing blindness,[13] sinusitis or nasal obstruction, odontogenic cyst, an oroantral fistula, other missing teeth, and sepsis.[13-15] Saleem et al[16] reported a 45-year-old man with 2-month history of hemoptysis. Litvin et al[17] reported a 57-year-old black woman with DC and an impacted tooth in orbital rim that presented with facial swelling. Kaygusuz et al[18] reported a mucocele associated with an ectopic tooth in the maxillary sinus, without fistula. Avitia et al[19] reported a case of orbital proptosis resulting from a DC in the maxillary sinus associated with a displaced tooth.

Bonder et al[20] studied 12 patients with teeth in the maxillary sinus by plain film radiography (PFR) and by CT with a dental software program. They found that CT was superior to PFR to determine proximity of the tooth to the sinus wall or its ankylosis, proper surgical planning, as well as prediction of prognosis or complications.

Ustuner et al[21] provided magnetic resonance imaging findings of the cystic lesion, which appears homogeneously hypointense on T1-weighted images and hyperintense on T2-weighted images. The impacted tooth appears hypointense on all sequences. Chronic secretions, air, and acute hemorrhage in the sinus may also appear hypointense, but their central location in the sinus distinguishes them from tooth, which lies eccentrically in the cyst wall.

On radiographic examination, DCs appear as unilocular radiolucencies of varying sizes, with well-defined sclerotic borders, associated with the crown of an unerupted tooth. If a follicular space on radiography is more than 5 mm, an odontogenic cyst can be suspected.[10]

Thus, CT scan is certainly superior to panoramic radiograph for the diagnosis of maxillary sinus pathology,[22] as plain radiography is limited by distortion and superimposition of anatomical structures. Better precision in localization of pathology with cross-sectional imaging resulted in more reliable diagnosis and aids treatment planning.[23] Both cone-beam CT and medical CT can be used for dental imaging as well as for three-dimensional reconstruction to aid surgical planning. Cone-beam CT has the advantages of good spatial resolution, substantially lower cost, smaller and lighter equipment, more convenient installation, and being easier to operate and maintain, and are generally more popular in the dental market. However, cone-beam CT has lower contrast resolution, limiting the discrimination between different tissue types. Also, CT is useful to delineate the three-dimensional morphology of the ectopic tooth, its inclination, and proximity to the sinus wall.[1,20]

The treatment of choice of ectopic teeth associated with the cystic lesion in maxillary sinus is surgical removal of the tooth along with enucleation of the cyst. In the literature, various techniques were mentioned, such as endoscopic procedure, extraoral approach, and intraoral removal.[24]

Di Pasquale and Shermetaro[8] used a nasal endoscope to create a middle meatal antrostomy and deliver the ectopic tooth and its cystic contents. The endoscopic techniques are being used for removal of an intranasal ectopic tooth obstructing nasal cavity and ectopic maxillary third molar obstructing osteomeatal complex.[6]

Endoscopic approach is associated with lesser operative as well as postoperative morbidity.[24] The danger of removing the tooth intraorally is that if any undue forces are exerted by instrument on the tooth, it will displace it into the infratemporal fossa. Hence, a careful removal is carried out in this case by the suction pressure. The tooth was pulled out by applying the tip of suction over the tooth and removed from the window after holding it with the help of "Ellis" forceps.[10]

In large cysts, an initial marsupialization to diminish the size of the osseous defect, followed by enucleation and tooth extraction, has been advocated. The major disadvantage is recurrence or persistence of the lesion.

In this case as the cyst was extended, a facial degloving was done which is only reported by Hunter;[24] obviously its advantages are sufficient surgical access and better control excision, so less chances of recurrence.

There is controversy regarding the management of large residual bony cavities with or without bone grafts. Numerous graft materials, such as autografts, allogenic, xenografts, and platelet-rich plasma are available for the same.[25]

In the present cases, relying on spontaneous bone regeneration, no bone graft was used. In fact, after 6 months, spontaneous filling of bone cavity was observed as with the studies of Shokier and Khalifa.[26]

Other than recurrence due to incomplete removal, complications related to this cyst are the potential to develop odontogenic tumors like ameloblastoma and malignancy like squamous cell carcinoma and mucoepidermoid carcinoma either from the lining epithelium which contains mucous-secreting cells or from the rests of odontogenic epithelium in the wall of the cyst.[6] Postoperative follow-up with radiographic examination at regular intervals is mandatory to rule out any recurrence.

CONCLUSION

Ectopic eruption into the nondentate region, such as maxillary sinus is rare and scantly documented. We report two cases of ectopic teeth in maxillary sinus associated to DC with their clinical features and surgical management. According to the literature, complete enucleation of the cyst and regular postoperative follow-up with radiographic examination is mandatory to rule out any recurrence. Nevertheless, no consensus about the efficacy technique is mentioned related to the position of the tooth or the size of the cyst.

REFERENCES

1. Lai YT, Luk YS, Fung KH. Anomalous morphology of an ectopic tooth in the maxillary sinus on three-dimensional computed tomography images. J Radiol Case Rep 2013 Feb;7(2):11-16.
2. Thakur G, Nair PP, Thomas S, Ahuja R, Kothari R. Dentigerous cyst associated with ectopic maxillary third molar in maxillary antrum. BMJ Case Rep 2011 May;2011:3873.
3. Srinivasa Prasad T, Sujatha G, Niazi TM, Rajesh P. Dentigerous cyst associated with an ectopic third molar in the maxillary sinus: a rare entity. Indian J Dent Res 2007 Jul-Sep;18(3):141-143.
4. Garde JB, Kulkarni AU, Dadhe DP. Ectopic tooth in the orbital floor: an unusual case of dentigerous cyst. BMJ Case Rep 2012 Jan;2012:5266.
5. Buyukkurt MC, Omezli MM, Miloglu O. Dentigerous cyst associated with an ectopic tooth in the maxillary sinus: a report of 3 cases and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2010 Jan;109(1):67-71.
6. Ramanojam S, Halli R, Hebbale M, Bhardwaj S. Ectopic tooth in maxillary sinus: case series. Ann Maxillofac Surg 2013 Jan;3(1):89-92.
7. Szilágyi A, Keszthelyi G, Nagy G, Madléna M. Oral manifestations of patients with Turner syndrome. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2000 May;89(5):577-584.
8. Di Pasquale P, Shermetaro C. Endoscopic removal of a dentigerous cyst producing unilateral maxillary sinus opacification on computed tomography. Ear Nose Throat J 2006 Nov;85(11):747-748.
9. Jones AV, Craig GT, Franklin CD. Range and demographics of odontogenic cysts diagnosed in a UK population over a 30-year period. J Oral Pathol Med 2006 Sep;35(8):500-507.

10. Kasat VO, Karjodkar FR, Laddha RS. Dentigerous cyst associated with an ectopic third molar in the maxillary sinus: a case report and review of literature. Contemp Clin Dent 2012 Jul-Sep;3(3):373-376.
11. Beriat GK, Beriat NC, Yalcinkaya E. Ectopic molar tooth in the maxillary sinus: a case report. Clin Dent Res 2011 Apr;35(2):35-40.
12. Abdollahifakhim S, Mousaviagdas M. Ectopic molar with maxillary sinus drainage obstruction and oroantral fistula. Iran J Otorhinolaryngol 2013 Jun;25(72):187-192.
13. Lamb JF, Husein OF, Spiess AC. Ectopic molar in the maxillary sinus precipitating a mucocele: a case report and literature review. Ear Nose Throat J 2009 Aug;88(8):E6-E11.
14. Dagistan S, Cakur B, Göregen M. A dentigerous cyst containing an ectopic canine tooth below the floor of the maxillary sinus: a case report. J Oral Sci 2007 Sep;49(3):249-252.
15. Erkmen N, Olmez S, Onerci M. Supernumerary tooth in the maxillary sinus: case report. Aust Dent J 1998 Dec;43(6):385-386.
16. Saleem T, Khalid V, Hameed A, Ghaffar S. Supernumerary, ectopic tooth in the maxillary antrum presenting with recurrent haemoptysis. Head Face Med 2010 Nov;6:26.
17. Litvin M, Caprice D, Infranco L. Dentigerous cyst of the maxilla with impacted tooth displaced into orbital rim and floor. Ear Nose Throat J 2008 Mar;87(3):160-162.
18. Kaygusuz I, Karlidag T, Akyigit A, Yalcin S. Mucocele associated with an ectopic tooth in the maxillary sinus: a case report. Int J Pediatr Otorhinolaryngol Extra 2011;6(4):401-402.
19. Avitia S, Hamilton JS, Osborne RF. Dentigerous cyst presenting as orbital proptosis. Ear Nose Throat J 2007 Jan;86(1): 23-24.
20. Bonder L, Tovi F, Bar-Ziv J. Teeth in the maxillary sinus–imaging and management. J Laryngol Otol 1997 Sep;111(9): 820-824.
21. Ustuner E, Fitoz S, Atasoy C, Erden I, Akyar S. Bilateral maxillary dentigerous cysts: a case report. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2003 May;95(5):632-635.
22. Maestre-Ferrín L, Galán-Gil S, Carrillo-García C, Peñarrocha-Diago M. Radiographic findings in the maxillary sinus: comparison of panoramic radiography with computed tomography. Int J Oral Maxillofac Implants 2011 Mar-Apr;26(2):341-346.
23. Botticelli S, Verna C, Cattaneo PM, Heidmann J, Melsen B. Two-versus three-dimensional imaging in subjects with unerupted maxillary canines. Eur J Orthod 2011 Aug;33(4):344-349.
24. Rai A, Rai NJ, Rai MA, Jain G. Transoral removal of ectopic maxillary third molar situated superiorly to maxillary antrum and posteroinferiorly to the floor of orbit. Indian J Dent Res 2013 Nov-Dec;24(6):756-758.
25. Ramakrishna A, Lambade P. Dentigerous cyst associated with ectopic canine and a supernumerary tooth: a rare occurrence. J Surg Tech Case Rep 2013 Jul;5(2):85-88.
26. Shokier HM, Khalifa GA. Assessment of bone healing in large bony defects after enucleation of jaw cysts without using any graft material using direct digital radiography and CT scan. C. D. J. 2009 Jan;25(1):35-42.