Case Report

Ectopic Tooth with Odontogenic Keratocyst in Myasthenia Gravis: A Rare Coexistence

Abstract

Ectopic tooth in nondentate areas is uncommon. Odontogenic keratocyst arising from impacted tooth in the inferior border of orbit is very rare. This article reports the case of a 22-year-old female with odontogenic keratocyst arising from an impacted left maxillary third molar in the inferior border of the orbit causing symptoms of chronic sinusitis. The patient also had myasthenia gravis which confounded the diagnosis. The cyst was surgically enucleated along with the tooth which relieved the symptoms of the patient.

Keywords: *Chronic sinusitis, ectopic tooth, impacted maxillary third molar, odontogenic keratocyst cyst*

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Introduction

Ectopic teeth are those that are impacted in unusual positions and displaced from their normal anatomic location.[1] Ectopic tooth are uncommon in nondentate areas, although reported in various sites including palate, orbit, nasal cavity, maxillary sinus, and chin.[2] The etiology may be due to trauma, infection, developmental disorders, or idiopathic.[3] Odontogenic keratocyst (OKC) is the third common odontogenic cyst that derives from odontogenic epithelial remnants in jaws.[4] It occurs in the 2nd–3rd decades of life and may go unnoticed throughout a patient's entire life or may turn aggressive and cause the expansion of jaws. In other instances, the patient becomes symptomatic and experiences the signs of sinus disease.[2] OKC involving maxillary sinus is extremely rare, with less than 1% of cases of all OKC occur in the maxilla involving sinus.[5] This is a case report describing an ectopic maxillary third molar in the inferior border of the orbit associated with OKC involving the maxillary sinus and causing the symptoms of recurrent chronic sinusitis.

Case Report

A 22-year-old female patient reported to the department of oral medicine with a history of pain and swelling in the left side of the face with occasional discharge of pus from the third molar region for the past 3 days. She gave a history of recurrent facial pain, heaviness of the head, and occasional pus discharge through the nose for the past 3 months. She gave a medical history of having myasthenia gravis from the age of 2 and being under regular medication (neostigmine 2 mg) for the past 20 years. The patient has undergone orthodontic treatment for the correction of malocclusion. Myofunctional appliance (twin block appliance) has been given for 1 year. On examination, there was a mild facial asymmetry caused by the swelling over the left maxillary region [Figure 1]. The mass was firm and tender on palpation. On intraoral examination, expansion of the left buccal cortex was seen obliterating the left buccal vestibule with the absence of upper left third molar. Mucosa over that area was erythematous and tender. No pus discharge was seen. Orthopantomogram (OPG) revealed impacted maxillary third molar in the infraorbital margin with haziness in the left maxillary sinus [Figure 2]. Computed tomographic image showed a well-defined radiolucency around the unerupted tooth seen along the roof of left maxillary antrum and extending into maxillary antrum measuring 3.20 cm × 3.10 cm with impending oroantral fistula. There was a complete opacification of maxillary sinus [Figure 3a and b]. Thinning of the medial wall and breach in the posterolateral

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Figure 1: Facial asymmetry on the left side due to a swelling in zygomatic region

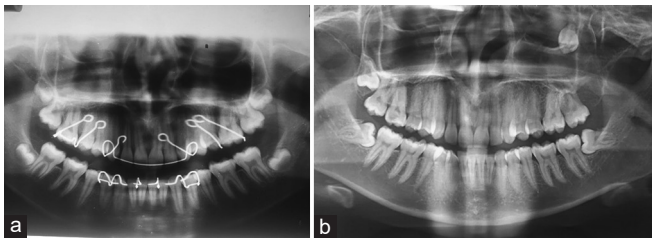

Figure 2: (a) Orthopantomogram taken during 12 years of age showing third molar tooth developing in normal position and (b) orthopantomogram taken presently shows the ectopic third molar in the inferior border of orbit and haziness in the left maxillary sinus

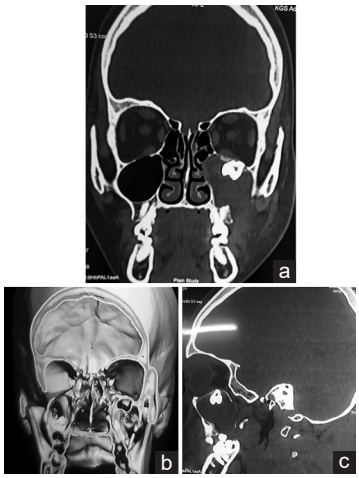

Figure 3: (a) Coronal section of computed tomographic image showing ectopic tooth in infraorbital margin close to inferior rectus muscle, complete opacification of left maxillary sinus, and thinning of its medial wall, (b) 3D reconstructed coronal view shows a well-defined radiolucency encircling the crown of ectopic tooth, (c) sagittal view showing breach in the posterolateral wall of maxillary sinus

wall of maxillary sinus was seen [Figure 3a and c]. The ectopic tooth was seen in the close proximity of inferior rectus muscle. A provisional diagnosis of dentigerous cyst in impacted maxillary molar was made with the list of differentials such as ameloblastoma, ameloblastic fibro-odontoma, and odontogenic keratocyst. The cyst was enucleated along with the maxillary third molar by Caldwell Luc procedure [Figure 4a and b]. The histopatholgical picture showed a fibrocollagenous cyst wall comprised stratified squamous epithelium with patchy chronic inflammatory cell infiltrate. The basal cells are arranged in the palisade pattern and were consistent with the features of odontogenic cyst [Figure 4c]. The patient had complete remission of symptoms and is on follow-up with no signs of sinusitis for the past 6 months.

Discussion

Ectopic teeth in dentate regions are common but unusual in nondentate areas. Rarely, ectopic impacted teeth have been found in mandibular condyle, sigmoid notch, nasal cavity, and maxillary sinus. Ectopic eruption of the third molar along the infraorbital margin is very rare and has been reported only in ten instances. In this patient, OPG taken at the age of twelve shows the developing crown in the normal position [Figure 2a]. The patient was lost to follow-up since she did not develop any malocclusion further. The reason for the migration of the developing tooth from its normal position to the infraorbital margin was enigmatic since no trauma or infection was reported. However, with the help of computed tomography, it was clear that OKC which has formed around the developing tooth has undergone expansion exerting pressure pushing the tooth up to the inferior border of orbit. The origin of OKC could be due to the entrapment of odontogenic epithelium of the developing tooth within the sinus due to its proximity to sinus floor.[5]

OKC has male predilection with the common occurrence in posterior mandible, ramus, and canine region in the maxilla followed by the maxillary tuberosity region. Maxilla is involved in 23.5% cases of OKCs.[6] Our patient is a female with an uncommon occurrence in maxillary sinus region.

Dentigerous cyst is the common type of developmental benign odontogenic cysts associated with an impacted tooth which is derived from the remnants of odontogenic epithelial cells.[7] However, our case is an OKC which is not common in order of differential diagnosis of cyst associated with an impacted teeth. OKCs usually occupy the maxillary sinus partially or totally and cause symptoms.[5] Our patient complained of chronic sinusitis with symptoms such as heaviness of head, facial pain, and pus discharge through the oral cavity for 6 months without any relief with conservative measures. An OPG and computed tomogram showed the complete opacification of the sinus. Moreover, this was a case of odontogenic sinusitis.[8] The presence of a cyst associated with an ectopic tooth along the infraorbital margin may cause visual disturbances due to optic nerve involvement due to resorption of the bone as reported by Demirtas *et al*.[3] In our patient, the ectopic tooth is present in close proximity to inferior rectus muscle. This patient is having myasthenia gravis causing mild ptosis of

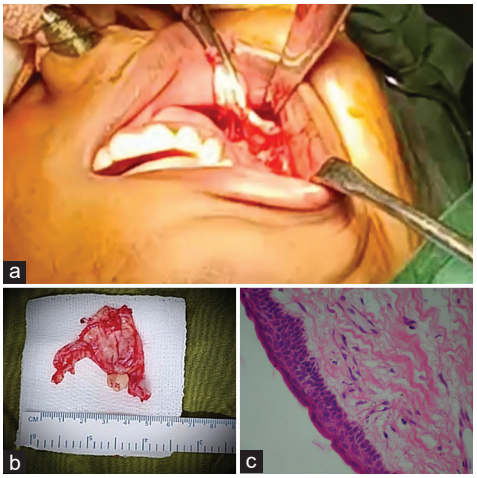

Figure 4: (a) Surgical removal of the ectopic tooth with cystic lining and contents by Caldwell luc procedure, (b) surgically removed ectopic tooth with cyst, (c) low power histopathological picture showing palisade pattern of basal cells with fibrocollagenous cystic wall

left eye and under daily medication of 2 mg neostigmine. This caused confusion as to whether the ectopic tooth is aggravating the existing ptosis. However, in our patient, the cyst developed inferiorly into the maxillary sinus not causing erosion of the inferior border of orbit and thus ophthalmic complications due to OKC in ectopic tooth is unlikely. This is the first case reported to have an OKC in an unusual position in a myasthenia gravis patient. Due to the propensity of recurrence of the cyst and its location in a strategic position which may impact her vision in future, the patient warrants lifelong surveillance by both clinical examination and three-dimensional imaging.

Conclusion

This case is reported for its rarity and to stress the importance of regular follow-up for any impacted tooth which may slowly undergo cystic transformation and may involve vital structures. Another learning point is that if chronic sinusitis is not relieved with conservative measures, a thorough clinical and radiological examination should be employed to find out odontogenic cause as seen in this case.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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