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Case Report

Dentigerous cyst associated with ectopic tooth at the roof of maxillary sinus

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Dentigerous cyst is the most common developmental odontogenic cyst of the jaws. They are frequently associated with impacted mandibular third molars and the maxillary canines. Association of ectopic tooth in the maxillary sinus with dentigerous cyst is quite uncommon. We report a case of dentigerous cyst associated with ectopic tooth at the roof of the maxillary sinus manifesting as a unilateral facial swelling with encroachment into the nose and orbit. This article addresses its unusual location, the important aspect of its etiopathologenesis, its clinical characteristics, differential diagnosis and management.

Key words: Dentigerous cyst, ectopic tooth, maxillary sinus, unilateral facial swelling, benign tumors.

INTRODUCTION

Dentigerous cyst is the most common developmental odontogenic cyst of the jaws and is frequently noted as an incidental finding on routine radiographs. The most common teeth affected are impacted mandibular third molars and permanent maxillary canines (Neville et al., 2002). The presence of ectopic tooth has been estimated to occur in 1% of the general population and has been found in the nasal cavity, maxillary sinus, mandibular bone, palate and orbital cavity of some patients (Erkmen et al., 1998, Thor, 2002, Wang et al., 2004).

Patients with dentigerous cyst in maxillary bone or sinus may present with facial swelling, purulent rhinorrhoea, nasal obstruction, external nasal deformity and epiphora. Radiographically, it typically shows a unilocular radiolucent shadow with a well-defined sclerotic border associated with the crown of an unerupted tooth (Wood and Goaz, 1997, White and Pharoah 2009).The standard treatment for an ectopic tooth is extraction of the tooth. However, dentigrous cyst associated with ectopic teeth within the maxillary sinus is often easily removed via a Caldwell-Luc procedure (Litvin et al., 2008, Goh, 2001, Atlas et al., 1997).

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We are aware that literature is awash with reported cases of dentigerous cysts with ectopic maxillary tooth (Takagi and Koyama, 1998, Ustuner et al., 2003, Srinivasa et al., 2007, Ngamdu et al., 2012) but to the best of our knowledge this is the first reported case of a dentigerous cyst associated with ectopic tooth at the roof of maxillary sinus in Port Harcourt, Rivers State, Nigeria. Therefore, this paper addresses its unusual location, the important aspect of its etiopathologenesis, its clinical characteristics, differential diagnosis and management

CASE REPORT

A 23 year-old house wife presented to our Ear Nose and Throat (ENT) outpatient clinic in the University of Port Harcourt Teaching Hospital (UPTH) Port Harcourt, Rivers State, Nigeria, with left nasal obstruction and a progressively increasing swelling in the left maxillary region of 5 years duration. No rhinorrhoea, epistaxis, otological and throat symptoms was observed. The medical history was non contributory; she does not smoke or drink alcohol, no history of undue exposure to chemicals, wood dust and irradiations.

The examination of the face revealed a left mid facial swelling that displaced the nose to the right side and has encroached into the left orbit (Figure 1). The mass was

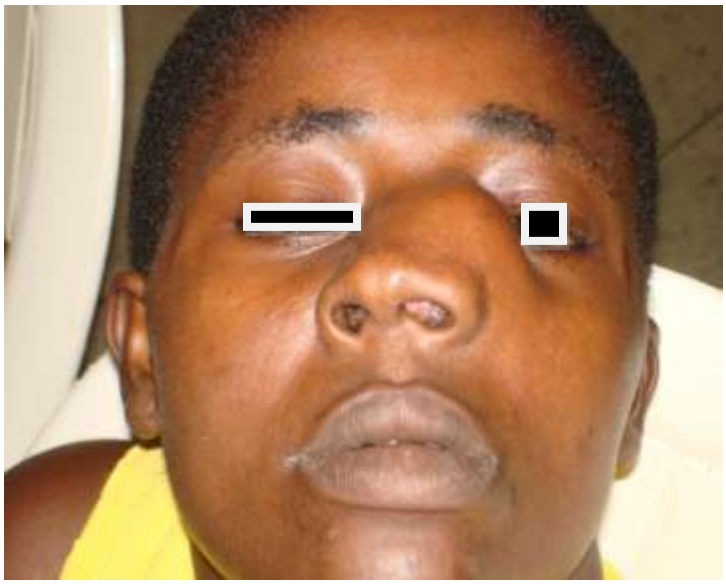

Figure 1. Patient's mid facial swelling at presentation to the ENT clinic.

Figure 2. Intra-operative findings (ectopic tooth at the roof of the left maxillary sinus after excision of cystic mass).

hard and non tender. On examination of the eyes there was diminished ocular vision of the left eye with excessive tearing. On examination of the nose, there was distortion of the nasal pyramid, deviation of the nasal septum to the right and a firm to hard left intranasal swelling arising from the lateral wall that was sensitive to touch but no contact bleeding. On examination of the oral cavity, all the permanent teeth were present and there were no abnormal intraoral findings. Post nasal space and indirect laryngoscopy examinations revealed normal findings. There were no associated neck lymph nodes and examinations of the systems appeared normal.

The radiograph of the nose and para nasal sinuses revealed soft tissue shadow in the left nasal cavity and opacification of the left maxillary antrum especially around the roof. There was also haziness of left ethmoidal air cells and frontal sinus. Chest radiograph was normal. The results of hematological and biochemical investigations were all within normal limits. Based on the above history, clinical findings and investigations, we reached a provisional diagnosis of a benign Sino nasal tumor and the patient was immediately worked up for medial maxillectomy combining a lateral rhinotomy incision with a lip splitting technique to help us gain better access to remove the tumour.

Figure 3. Patient on the 5th day post operatively with stitches insitu.

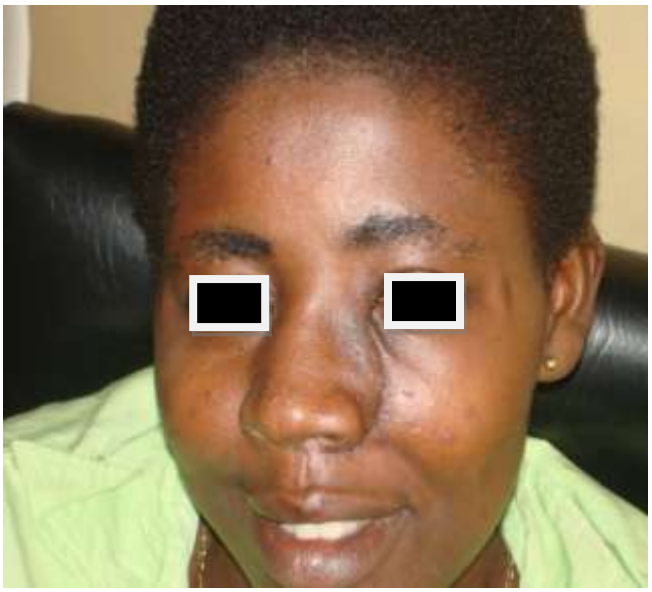

Figure 4. Patient's face 1 month post operatively.

General anaesthesia with endotracheal intubation and adequate muscle relaxation was used for the procedure. Intra-operative findings were that of a cystic mass that encroached into the left nasal cavity harboring an ectopic tooth at the roof of the maxillary sinus. The entire cystic sac was excised and submitted along with the tooth for histopathology examination which later showed that the cystic mass had connective tissue capsule made up of thin delicate collagen arranged parallel to each other. Inflammatory cells were observed and the lining epithelium was made up of layers of flat cells resembling reduced enamel epithelium confirming dentigerous cyst and a canine tooth. Figure 2 shows the ectopic tooth attached to the roof of the maxillary sinus. Post operative management of the patient was uneventful and patient was discharged home on the 7th day post operatively to continue follow up visits on outpatient basis. Figure 3 and 4 show patient on the 5th day and 1 month post operatively. Patient was followed up for 5 years and there was no reoccurrence.

DISCUSSION

Dentigerous cyst is formed by the hydrostatic force exerted by the accumulation of fluid between reduced

enamel epithelium and the tooth crown of unerupted tooth. As such the cyst encloses the crown and is attached at the neck at the cemento-enamel junction (Ikeshima and Tamura, 2002). They almost exclusively occur in permanent dentition and the cyst is lined by stratified squamous nonkeratinizing epithelium. The reduced enamel epithelium surrounds a developing tooth and degenerates as a tooth is erupting.

Dentigerous cysts are usually diagnosed during routine plain radiographic examination. Usually, it shows a well-defined radiolucency with sclerotic border associated with the crown of an ectopic tooth (Ikeshima and Tamura, 2002).

However, this picture was absent in the radiograph of our patient and as such, the diagnosis could not be suspected radiologically with plain radiographs of the nose and paranasal sinuses prior to surgical intervention. Unfortunately, the patient was unable to afford a computerized tomography scan of the nose and paranasal sinuses which would have shown clearly the features of dentigerous cyst and the ectopic tooth at the roof of the maxillary sinus as revealed intra-operatively.

Typically, dentigerous cysts are painless and small. However, they may be large and result in a palpable mass as seen in our patient. It usually presents in the second or third decade of life and can originate from any tooth including supernumerary tooth (Goh, 2001). Furthermore, it can be found in children and shows a male predilection (Shear, 1983, Kalaskar et al., 2007). However, in our case the patient was a female in the second decade of life. Clinical presentation may include facial swelling, intra oral swelling, nasal symptoms and ophthalmological manifestations of nose and paranasal sinus diseases (Onotai and Ejimadu, 2013).

The differential diagnosis of dentigerous cyst includes odontogenic tumor, unicystic ameloblastoma, fibrous dysplasia of the maxilla, osteoma of the maxilla and sino nasal malignant tumors (Hall et al., 1990, Minami et al., 1992). When small, it is difficult to differentiate a dentigerous cyst from a large but normal dental follicle. Occasionally, other more ominous lesions arise within the walls of the dentigerous cyst, including mucoepidermoid carcinoma arising from mucous cells within the cyst walls and squamous cell carcinoma (Gulbranson et al., 2002).

Treatment usually involves removal of the entire cyst and the associated unerupted tooth. In patients with very large lesion or who are unfit medically, marsupialisaiton is an option. Our patient had removal of the entire cyst and the associated ectopic tooth using a lateral rhinotomy incision with a lip splitting technique. However, we would have used an intra-oral approach if we had clinched the diagnosis prior to surgery (Srinivasa et al., 2007). The major disadvantage of our approach was the poor cosmetic scar on the patient face even though it helped us gain better access as the cyst may reoccur if parts of its lining are left insitu. Meanwhile, our patient has no evidence of recurrence after being followed up for 5 years.

CONCLUSION

The peculiarity of this case borders on the unusual location of the dentigerous cyst and the diagnostic dilemma posed by this clinical condition as a result of lack of a superior radiological investigation. To determine the best treatment for patients it is important for clinicians to have a high index of suspicion and as well carry out comprehensive radiological assessments of patients suspected to have lesions that mimic Sino nasal tumors.

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