Reminder of important clinical lesson

Maxillary sinus swelling in a child: clinical dilemma

Rashmi Goyal,¹ Alok Kumar,¹ Dipti Saxena,² Rakesh Biswas³¹ Department of ENT, People's College of Medical Sciences and Research Centre, Bhanpur Bypass, Bhopal, India² Department of Oral and Maxillofacial Surgery, People's College of Medical Sciences and Research Centre, Bhopal, India³ Department of Medicine, People's College of Medical Sciences and Research Centre, Bhopal, India

Correspondence to Rakesh Biswas, rakesh7biswas@gmail.com

Summary

A 7-year-old girl had presented with right cheek swelling noted 3 months earlier. It was slowly progressive. Swelling was well defined, non-tender, smooth, firm in consistency and measured 4×4 cm. Intraoral examination revealed no missing teeth. Permanent teeth – central incisors (upper and lower), primary lateral incisor, canine and first and second molar were present. X-ray paranasal sinuses showed well-defined unilocular radiolucency with a thin sclerotic border enclosing permanent first molar, premolar and canine on the right side and first molar on the left side. The cyst was removed by enucleation procedure. Mucoperiosteal flap was elevated, and paper thin bone overlying the cyst was removed. The cyst was enclosing the permanent canine, premolar and molar teeth. Histopathological report was consistent with the dentigerous cyst. After 1-year follow-up, the patient has no obvious swelling over the cheek.

BACKGROUND

The case was important because bilateral non-syndromic dentigerous cyst is rarely encountered. Although mostly associated with mandibular dentition, in our case it was associated with bilateral maxillary dentition.

There is a male predilection of 1.6:1, but in our case the patient was a female. The swelling completely resolved after enucleation.

CASE PRESENTATION

A 7-year-old girl had presented with a history of a right cheek swelling noticed 3 months earlier. The swelling was slowly progressive. It was painless, there was no history of nasal blockage, nasal discharge, anosmia or symptoms of sinusitis. There was no history of trauma or other significant medical history. Swelling was well defined, non-tender, firm in consistency and measured about 4×4 cm. There were no signs of inflammation.

On intraoral examination, right upper first premolar (primary) was carious. There was swelling over the right upper premolar region extending to the canine region. The swelling was firm, hard in consistency, non-tender, smooth and well defined. There was no bruit or pulsation. The overlying mucosa was normal. Permanent teeth – central incisors (upper and lower) were present. Primary dentition – lateral incisors, canine and first and second molars were present. Posterior pharyngeal wall was normal with no postnasal discharge; there were no missing teeth. Nasal endoscopy was performed and it was normal.

INVESTIGATIONS

X-ray paranasal sinuses (PNS) showed a well-defined unilocular radiolucency with a thin sclerotic border, enclosing permanent first molar, premolar and canine on the right side and first molar on the left side (figure 1). CT-PNS confirmed the findings of the x-ray PNS (figure 2).

Aspiration of the lesion was performed through the canine fossa. The return fluid was straw coloured.

Histopathology of enucleated cyst was consistent with that of the dentigerous cyst.

DIFFERENTIAL DIAGNOSIS

A large periapical cyst, odontogenic keratocyst, central giant-cell granuloma and unicystic ameloblastoma can mimic a dentigerous cyst. Odontogenic keratocyst and unicystic ameloblastoma most frequently occur in the molar region of the lower jaw in the second and third decades of life. Odontogenic keratocyst (primordial cyst) is a cyst with keratinised epithelium. Primordial cyst forms from the tooth bud and grows instead of a tooth. The primordial cyst tends to extend in the medullary cavity and expansion of the cortex occurs late. A radiograph will not differentiate between a radiolucency associated with the root of a non-vital primary teeth and the crown of unerupted teeth.¹

TREATMENT

Under general anaesthesia, the oral cavity was exposed and incision was given over the right muco-buccal fold extending from central incisor (without injuring the frenulum) to first molar. Mucoperiosteal flap was elevated to expose the lesion. Paper thin bone was present covering the lesion, which was removed to expose the cyst. The cyst was finally separated from the bony cavity and taken out in toto. The cyst was enclosing a permanent canine, premolar and molar. The antrum was filled with Ab-gel and the mucoperiosteal flap was replaced.

The enucleated specimen clearly showed a well-defined cyst containing permanent canine, first molar and premolar teeth (figure 3). The enucleated cyst was sent for histopathological examination.

Figure 1 Paranasal sinuses (Water's view) showing bilateral dentigerous cyst enclosing the teeth.

Figure 2 CT scan: paranasal sinuses' coronal view showing bilateral dentigerous cyst.

Figure 3 Specimen of enucleated dentigerous cyst with right canine, premolar and molar.

OUTCOME AND FOLLOW-UP

The cystic lesion was enucleated from the bony cavity. There was no intraoperative and postoperative complication. Postoperative swelling had completely resolved. After 1-year follow-up, the face of the child is absolutely normal and her parents are satisfied with the cosmetic results.

DISCUSSION

A dentigerous cyst arises from the dental follicle of an unerupted or developing tooth. It is the second most common odontogenic cyst after the radicular cyst.¹ These cysts occur most frequently in mandibular third molar, and the other teeth that are commonly affected are, in the order of frequency, maxillary canine, maxillary third molar and, rarely, maxillary right central incisor.² It apparently develops by the accumulation of fluid between the reduced enamel epithelium and the tooth crown of an unerupted tooth. Peak incidence is in the teenage years and in the twenties. There is a male predilection of 1.6:1.¹

A dentigerous cyst may be discovered as an accidental radiographic finding or by the examination of a clinical expansion. Because this cyst can attain a very large size (10–15 cm), some present with bony expansion and facial fullness. The tooth from which it arises is clinically absent. Regardless of the degree of absorption or the cyst's size, inferior alveolar nerve sensation in the mandible or superior alveolar nerve plexus sensation in the maxilla is not altered unless a pathological fracture, or more rarely an infection, causes paresthesia.¹ In our case, the patient presents with facial fullness.

Dentigerous cysts are very common developmental cysts and they are generally solitary. Bilateral dentigerous cysts usually occur in association with syndromes like mucopolysaccharidosis (type VI) and cleidocranial dysplasia. Both diseases cause alteration in tooth development or in their eruption. Bilateral dentigerous cysts are extremely rare in the absence of a syndrome or systemic disease.³ In the present case report, patient had bilateral dentigerous cyst, which was not associated with any syndrome. Although the dentigerous cyst is more common in the mandibular jaw, in the present case the maxillary jaw was involved.

There are two leading theories on the formation of dentigerous cysts. The first begins with fluid accumulation between the reduced enamel epithelium and the crown of the tooth. The fluid pressure incites a proliferation of the reduced enamel epithelium into a cyst, which is attached to a cement enamel junction and includes the tooth crown as one of its boundaries. The other theory begins with a breakdown of the stellate reticulum, which forms a fluid between the inner and outer enamel epithelium. The fluid pressure incites a proliferation of the outer enamel epithelium, which remains attached to the tooth at the cemento-enamel junction; the inner enamel epithelium is then pressed onto the crown surface. In each theory, the fluid generates cystic proliferation by its hyperosmolar content created by cellular breakdown and cell products, causing an osmotic gradient to pump fluid into the cyst lumen.¹ Toller stated that the likely origin of dentigerous cyst is the breakdown of proliferating cells of the follicle after impeded eruption. These breakdown products result in increased osmotic tension and hence cyst formation.⁴

Dentigerous cysts most commonly occur in the second and third decades of life. These lesions can also be found in children and adolescents and show a male predilection, but our case was a 7-year-old girl. Studies have shown that the incidence rate of dentigerous cysts involving the maxillary central incisor was 1.5% as compared with 45.7% involving the mandibular third molar.⁵ In the present case report, the dentigerous cyst involved maxillary first molar, premolar and canine teeth. It is important to perform radiographic examinations in case of unerupted teeth. Initially, a panoramic radiograph may be used for this examination. However, in cases of extensive lesion, CT imaging becomes necessary.^{6,7} CT imaging gives information about origin, size, content, cortical plates and relationship of the lesion to adjacent anatomical structures.

A large periapical cyst, odontogenic keratocyst, central giant-cell granuloma and unicystic ameloblastoma can mimic a dentigerous cyst. Odontogenic keratocyst and unicystic ameloblastoma most frequently occur in the molar region of the lower jaws in the second and third decades of life. A radiograph will not differentiate between a radiolucency associated with the root of a non-vital primary teeth and the crown of uninterrupted teeth.⁸ Unlike other odontogenic cysts, the epithelial cells lining the lumen of the dentigerous cysts possess an unusual ability to undergo metaplastic transition. On occasion, some untreated dentigerous cysts rarely develop into an odontogenic tumour (eg, ameloblastoma) or a malignancy (eg, oral squamous cell carcinoma).⁹

Odontogenic keratocyst (primordial cyst) is a cyst with keratinised epithelium. Primordial cyst forms from the tooth bud. The primordial cyst tends to extend in the medullary cavity and expansion of the cortex occurs late.

The preferred treatment is a complete removal of the lesion from the bony cavity by enucleation as marsupialisation is less ideal; it runs the risk of allowing an ameloblastoma or other neoplastic transformation of the cyst lining to develop into a more invasive case. It also submits the

wound to a slower healing process, a more laborious post-operative course and a reduction in the final bone regeneration.¹ In the present case, enucleation of the cyst was done.

Learning points

- ▶ Cheek swelling in a child should be properly investigated.
- ▶ Developing teeth should be taken care of.
- ▶ Early surgical intervention prevents cosmetic deformity.

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Competing interests None.

Patient consent Obtained.

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