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CASE REPORT

Multiple unilateral maxillary dentigerous cysts in a non-syndromic patient: A case report and review of the literature

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Summary Dentigerous cysts are the most common developmental odontogenic cysts of the jaw, arising from impacted, embedded or unerupted teeth. Although mostly solitary, multiple cysts occur in association with syndromes such as mucopolysaccharidosis type VI, cleidocranial dysplasia, basal cell nevus syndrome, and Gardner's syndrome. Multiple cysts in non-syndromic patients are extremely rare, occurring almost exclusively in the mandible. We report the case of a four-year-old girl with painless right-sided facial swelling, diagnosed with two dentigerous cysts in the right maxillary sinus, who successfully underwent enucleation. To our knowledge this is the only case of multiple unilateral maxillary dentigerous cysts in a healthy, non-syndromic patient.

1. Introduction

Dentigerous cysts are benign odontogenic lesions arising from the completed crown of impacted, embedded or unerupted teeth, and are most common in the first two decades of life [1]. They are the most common developmental odontogenic cyst, and second most common cystic lesion of the jaws next to radicular cysts. Seventy-five percent of dentigerous cysts arise in the mandible [2]. Patients are typically asymptomatic, and when not detected early enough on dental radiography, they may progress to a considerable size with the patient presenting with painless facial swelling.

Dentigerous cysts are usually solitary, but are known to be multiple in patients with certain syndromes, such as mucopolysaccharidosis type VI and basal cell nevus syndrome, Gardner's syndrome and Cleidocranial dysplasia [2,3]. There are limited cases of multiple cysts occurring in non-syndromic

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patients. Dentigerous cysts are also known to occur in association with other pathologic odontogenic entities such as mucoepidermoid carcinoma, ameloblastoma and squamous cell carcinoma [4]. Alternatively, in rare instances, dentigerous cysts may themselves arise from supernumerary teeth, or even from odontomas [5].

This report describes a case of an otherwise healthy four-year-old non-syndromic girl who presented with two dentigerous cysts in the right maxillary sinus.

2. Case

A four-year-old girl was referred to the Department of Otolaryngology for a painless facial swelling involving the right cheek. The swelling had progressed in size over the course of 9 months. Three months prior to presentation, the patient developed right nasal obstructive symptoms accompanied by right ocular infections. There were no visual disturbances or paresthesias. There was no history of dental procedure or dental/gingival changes, nor was there any weight loss, fatigue, or generalized neurological symptoms. The patient maintained all of her cognitive and functional abilities and continued to thrive. She was in otherwise good health without any previous significant medical history or associated syndromes.

Physical examination revealed a swelling overlying the right maxillary sinus. There was no nasal deviation, nor was there any apparent ocular pathology. There were no skin changes and the area was non-tender (Fig. 1). Intranasal speculum examination revealed a bulge in the right lateral nasal wall causing obstruction. Oral examination revealed normal dentition without any gingival pathology or trismus. A full physical examination did not reveal any other findings.

Fig. 1 Pre-operative evaluation of the patient reveals a right-sided facial swelling overlying the right maxillary sinus.

2.1. Imaging

Computed tomography revealed two large cystic structures in the right maxillary region each associated with an isolated secondary tooth. There was a smaller anteromedial cyst causing protrusion of the anterior maxillary wall, with a larger lateral cyst deforming, but not invading the right orbital floor (Fig. 2). Magnetic resonance imaging revealed abnormal fluid signal intensity increasing in T2 and low in T1 with subtle rim enhancement and expansion/thinning of the right maxillary sinus. The right osteomeatal complex was completely obliterated without extension into the orbit (Fig. 3). The pantomogram did not aid in the diagnostic or therapeutic processes.

2.2. Management

Under general anesthesia, the patient underwent enucleation of the two right intramaxillary cysts and their respective teeth via a Caldwell-Luc approach and endoscopic sinus surgery (Fig. 4). Intraoperative findings revealed a third tooth in the maxillary sinus not associated with any cyst and was left in situ. The patient had an uneventful post-operative course and was discharged home the next day. Follow-up at 10 days showed good healing at the surgical sites without any paresthesias.

2.3. Pathology

Histologic analysis revealed the presence of two dentigerous cysts each arising from a formed secondary tooth. Presence of tissue consistent with compound odontoma was also present, but not deriving from or giving rise to these cysts (Fig. 5).

3. Discussion

Dentigerous, or *follicular* cysts, are the second most common odontogenic cysts of the jaws after the periapical or *radicular* cyst, and make up an estimated 14—20% of all jaw cysts [6]. They are mostly associated with impacted, embedded or unerupted teeth, and most commonly arise from mandibular third molars, followed by maxillary third molars and maxillary canines [4]. In 5% of cases, they arise from supernumerary teeth, and in limited cases can arise from odontomas [5]. They are more prevalent in