DENTIGEROUS CYST: BILATERAL PRESENTATION IN A 13 YR OLD

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ABSTRACT

Dentigerous cysts are the most common developmental cysts of the jaws, most frequently associated with impacted mandibular third molar teeth. Bilateral dentigerous cysts are rare and occur typically in association with a developmental syndrome. The reported occurrence of bilateral dentigerous cysts in the absence of a syndrome is rare and, to date, only 11 cases have been described. Here, we report a case of bilateral non-syndromic, dentigerous cysts and review the literature for this unusual finding.

Keywords: Dentigerous cyst, Female, Young, Bilateral

INTRODUCTION

A dentigerous cyst is an epithelial-lined developmental cavity that encloses the crown of an unerupted tooth at the cementoenamel junction. Dentigerous cysts are the second most common odontogenic cysts after radicular cysts, accounting for approximately 24% of all true cysts in the jaws[1]. Their frequency in the general population has been estimated at 1.44 cyst for every 100 unerupted teeth[2]. The cyst arises from the separation of the follicle from the crown of an unerupted tooth, and although it may involve any tooth, the mandibular third molars are the most commonly affected.

Dentigerous cysts are frequently discovered when radiographs are taken to investigate a failure of tooth eruption, a missing tooth or malalignment. There is usually no pain or discomfort associated with the cyst unless it becomes secondarily infected. Radiographs show a unilocular, radiolucent lesion characterized by well-defined sclerotic margins and associated with the crown of an unerupted tooth. While a normal follicular space is 3 to 4 mm, a dentigerous cyst can be suspected when the space is more than 5 mm[3].

Most dentigerous cysts are solitary. Bilateral and multiple cysts are usually found in association with a number of syndromes including cleidocranial dysplasia and Maroteaux-Lamy syndrome[4]. In the absence of these syndromes, bilateral dentigerous cysts associated with the mandibular third molars are rare. To date, there have been only 11 reported cases in the literature (Table I)[5-15]. Here, we report the unusual occurrence of nonsyndromic bilateral dentigerous cysts associated with a mandibular premolar.

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Authors	Year	Sex	Race	Age (yrs.)	Location	Treatment
Sands and Tocchio5	1998	F	N/A	3	Md. central incisors and first molars	Enucleation
Banderas and others6	1996	M	C	38	Md. third molars	Enucleation
O'Neil and others7	1989	M	Bl	5	Md. first molars	Enucleation
Eidinger8	1989	M	C	15	Md. first molars	Enucleation
McDonnell9	1988	M	N/A	15	Md. second premolar and second molar teeth	Enucleation
Crinzi10	1982	F	Bl	15	Md. third molars	Enucleation
Swerdloff and others11	1980	F	C	7	Md. first molars	Enucleation
Burton and others12	1980	F	Bl	57	Md. third molars	Enucleation
Callaghan13	1973	M	C	38	Md. third molars	Enucleation
Stanback14	1970	M	N/A	9	Md. first molars	Enucleation
Myers15	1943	F	N/A	19	Md. third molars	Enucleation

N/A= Not available M = male C = Caucasian F = female Bl = Black Md. = mandibular

CASE REPORT

A 13year-old Nepali girl was referred to the department of dentistry of College of Medical Sciences, by an outside dental service for the evaluation of an asymptomatic, lesion in the right mandible. Intraoral examination revealed malocclusion in her dentition and clinically absent right mandibular first premolar teeth. No extraoral swellings or tenderness in relation to the mandibular premolar was noted. The patient's medical history was non-significant. There were no associated syndromes present. However, she had Dolichocephalism and was sent for examination and opinion to the Dept. of Paedriatics. However, no other skeletal, biochemical or haematological parameters suggested her having a syndrome.

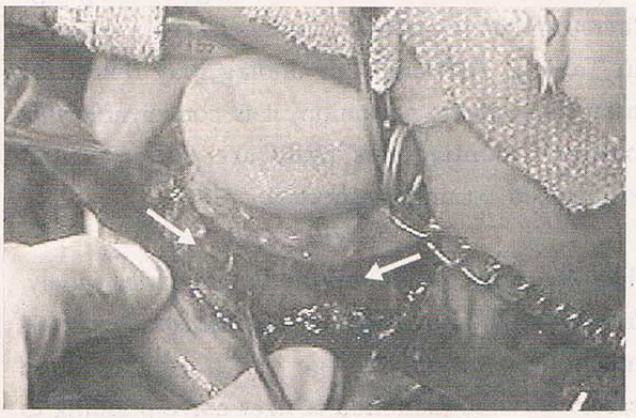

Fig.I: Bilateral dentigerous cysts of anterior mandible. The periosteal elevator is pointing at the midline, where there is sound bone. White arrows point towards the cysts.

RADIOGRAPHIC FINDINGS

Lateral Mandible radiograph showed impacted right first premolar molar. AnteroPosterior view of Mandible revealed bilateral, unilocular well-defined corticated radiolucency extending from the impacted premolar to the contralateral side, with intact cortical bone in the central incisor region.

TREATMENT

The clinical diagnosis was of a bilaterally extending dentigerous cyst. Under general anesthesia, a labial vestibular flap was raised and the cysts enucleated together with the associated right premolar tooth. The left premolar tooth was erupting in the oral cavity and hence was left as it is. The cortical bone below the central incisiors was guttered to connect the bilaterally extending cyst. The flap edges were approximated, leaving a 2 X 2 cm gap in the midline, for postoperative irrigation and observation. Healing was uneventful, and one

week after the operation, the surgical sites showed good healing. There was partial paresthesia on the right side affecting the mental nerve distribution of the inferior alveolar nerve, but after three weeks, there had been complete healing. There was also no evidence of recurrence of the cysts, as the cavity size shrunk and an acrylic obturator was provided.

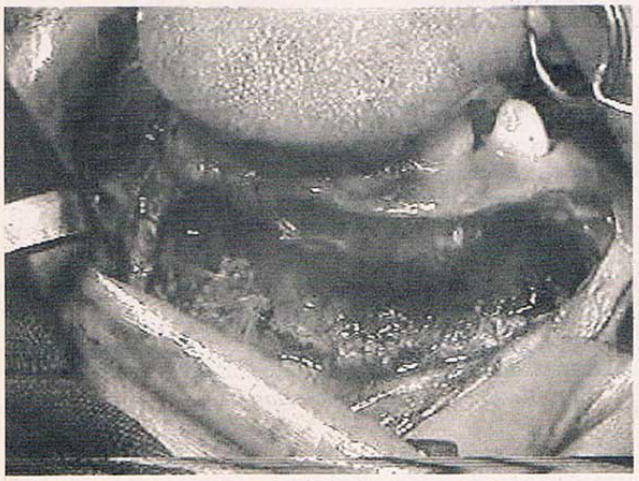

Fig. 2: Enucleation of the cysts with residual void is seen in this picture.

PATHOLOGY

The submitted specimen consisted of two sacs of soft tissue, the largest measuring 30 X 5 X 7 mm. Microscopic sections of both specimens were similar, showing cyst walls composed of fibrous tissue and lined by stratified squamous, non-keratinized epithelium with Rushton bodies (Fig.3).

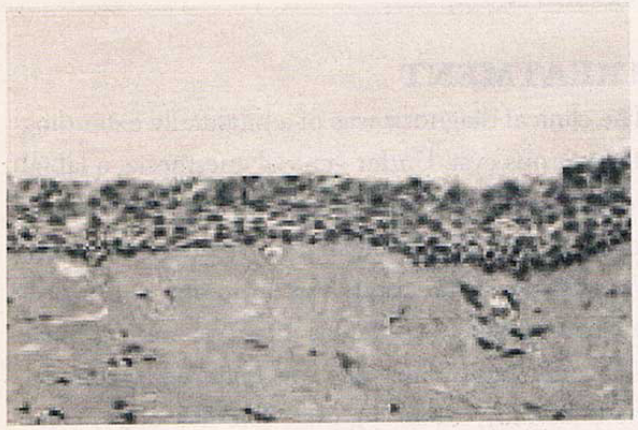

Fig. 3: Photomicrograph showingcyst wall composed offibrous tissue and lined by non-keratinized stratified sqaumous epithelium. (H& E X 40)

DISCUSSION

Although dentigerous cysts are common developmental cysts, reported bilateral dentigerous cysts are extremely rare. Bilateral or multiple dentigerous cysts are usually associated with the Maroteaux-Lamy (mucopolysaccharidosis, type VI) syndrome[16] and cleidocranial dysplasia[17]. Both are developmental conditions that are detected in young individuals with stigmata of the syndromes.

Maroteaux-Lamy syndrome is one of the mucopolysaccharidoses (MPS) group of diseases resulting from a genetic defect in the degradation of specific mucopolysaccharides. With this syndrome, there is a deficiency of N-acetyl-4-sulphatase that results in impaired degradation of dermatan sulphate, which accumulates in tissues and is excreted in the urine. Dental features include unerupted dentition, dentigerous cysts, malocclusions, condylar defects, and gingival hyperplasia[16]. Cleidocranial dysplasia is an autosomal dominantly inherited disorder that results in a partial or complete absence of clavicles, short stature, frontal and parietal bossing, maxillary micrognathia, prolonged retention of the primary dentition, delayed eruption of the permanent dentition, and unerupted supernumerary teeth.[17] Multiple dentigerous cyst formation occurs in both conditions and can develop at any site in the upper or lower jaws.

Bilateral dentigerous cysts are rare in the absence of an underlying syndrome or systemic disease. An extensive search of the English language literature has identified only the 11 cases reported in Table I. Although this finding may reflect the true rarity of the condition, it is conceivable that bilateral dentigerous cysts are either under-recognized or under-reported.

The age range for the reported cases varies widely, from 5 to 57 years of age. Four of the cysts occurred in children under the age of 12. All but three cases[6,12,13] were identified at ages corresponding to the normal eruption times of the affected teeth. As with our case, these three cases occurred in

asymptomatic individuals, which accounts for the delayed diagnosis. All but two of the 11 cases[5,9] have been associated with mandibular molar teeth, with five of these associated with the third molar teeth[6,10-13]. There have been no reported cases of nonsyndromic, bilateral dentigerous cysts occurring in all four dental quadrants.

Since cysts can attain considerable size with minimal or no symptoms, early detection and removal of the cysts is important to reduce morbidity. Moreover, all but one[11] of the reported cases, including ours, presented without pain. And, all but one were discovered during investigation of asymptomatic slow-growing swellings.[11]

It is therefore important to perform radiographic examination of all unerupted teeth. While bite-wing and periapical radiography is typically performed in the routine examination of patients with a healthy dentition, this series of radiographs may occasionally fail to delineate the full extent of a lesion if present. A panoramic radiograph supplemented with skull series or more advanced imaging such as tomography may permit a better delineation of the extent of the lesion and its relationship to adjacent anatomical structures.

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