Coincidental Overlapping Cystic Pathological Changes: A Case against Marsupialisation

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Abstract It is relatively common to observe a solitary well defined periapical cystic change associated with decayed primary tooth displacing the underlying developing permanent tooth bud. Marsupialisation of such cysts is indicated so as to allow for eruption of the underlying tooth bud. A similar solitary well defined periapical cystic lesion involving the mixed dentition in a child was planned for enucleation. Surprisingly, during the surgical intervention two distinct separate cystic pathological changes were observed. Marsupialisation of the cystic defect with extraction of the primary tooth would have left the enlarged follicle with the developing tooth bud unaddressed. The pathological process associated with the dilated follicle would continue to progress further and may require an additional surgical intervention. The distinction between enlarged follicle and dentigerous cyst clinically is also important. Considering the size, extent and presence of the large bony cavity during the surgical intervention enucleation of both the pathological changes was performed.

Keywords Radicular cyst · Enlarged follicle · Marsupialisation

Introduction

There are numerous literature reports of various pathological changes developing in the wall of odontogenic

cysts [1, 2], but coexisting overlapping cystic pathological changes within the same anatomic zone involving a radicular cyst from primary dentition and enlarged follicle with developing tooth bud from the permanent dentition, not very well delineated on the radiographs, have according to the authors knowledge not been reported. Marsupialisation of the cyst may be appropriate if it is hoped that the tooth will erupt. This will allow free drainage of the liquid content of the cyst, shrinkage of the cyst sac, and new bone formation on its capsular aspect [3]. Association between infected primary teeth and the formation of dentigerous cysts has been observed previously. Azaz and Shteyer [4] described five dentigerous cysts involving lower second premolars and associated with non vital predecessors. Radiographically the follicle appears as a homogeneous radiolucent space around the tooth with a thin outer radiopaque border. Since cystic change can occur in these follicles, it is important to identify any developing pathology at an early stage. A pericoronal space of greater than 2.5 mm on an intraoral radiograph and greater than 3 mm on a rotational panoramic radiograph should be regarded as suspicious [5].

Case Report

A 10 year old boy presented with a complaint of swelling on the right lower jaw from the past 2–3 months. There was a history of pain in the lower right back tooth region 6 months back which subsided on taking antibiotics and analgesics prescribed by the local dentist. On examination, there was a mild diffuse swelling over the right lower jaw. Intra orally the lower right deciduous second molar was grossly decayed with swelling obliterating the right lower buccal vestibule. There was a remarkable expansion of the

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buccal cortical plate. Swelling was tender, soft to firm in consistency and fluctuant in areas where egg shell crackling was elicitable. There were no neurosensory deficits. Lower right E was non vital. OPG revealed a solitary, well defined periapical radiolucency, approximately 2 cm by 1.5 cm in anteroposterior and superio-inferior dimensions, associated with the roots of the deciduous second molar tooth extending to involve the underlying tooth bud of premolar tooth (Fig. 1). The tooth bud appeared to be displaced towards the lower border of the mandible. Aspiration revealed a yellowish straw colored fluid. A clinical diagnosis of a radicular cyst involving the decayed deciduous molar was made. The possibility of a dentigerous cyst originating from tooth bud of first premolar and involving the deciduous second molar was also analyzed. Extraction of the decayed primary tooth and marsupialisation of the cystic cavity was considered as an option but a decision was made to enucleate the cystic lesion with the associated tooth bud under G.A, as the predictable eruption pattern of the impacted tooth was questionable from the orthodontic perspective.

Under G.A, mucoperiosteal flap was raised and the marked expansion of the buccal cortical plate was evident (Fig. 2). The cystic lesion was enucleated in toto expecting the tooth bud to be contained within the cavity. On examining the enucleated specimen the tooth bud did not seem in situ. The large bony cavity was explored in the body of the mandible. Surprisingly another distinct lining attached to the primary cystic cavity was observed concealed towards the mesial side under the distal aspect of lower right D. The tooth bud of the unerupted premolar was displaced lingually along the lower border of the mandible and was contained within this separate cystic cavity. This lining containing the tooth bud was carefully enucleated in

Fig. 1 Pre-op OPG X-ray showing a solitary well defined radiolucency associated with lower right E displacing the tooth bud of second premolar towards the lower border

Fig. 2 Marked buccal expansion

Fig. 3 Cystic defects

toto taking care so as to preserve the transition zone attaching both the cystic lesions. Distinct bony cavities were identifiable after enucleation (Fig. 3).

The larger cystic lesion approximately 3×2 cm, was dark red in colour being attached to the roots of the deciduous molar whereas the cystic lining with the impacted premolar approximately 1.5×1 cm, was clinically of smoother texture, thicker and bright pink in appearance (Fig. 4). The difference in the appearance of these pathological entities was distinctly appreciable on visual examination, the transition zone being the area where both lesions were joined together. The attached cystic specimens were sent for histopathological examination.

The histopathology report of tissue attached to the grossly decayed deciduous molar was suggestive of a radicular cyst (Fig. 5), whereas the tissue surrounding the impacted premolar tooth bud was reported to be an enlarged follicle (Fig. 6).

Fig. 4 Attached cystic pathologies

Fig. 5 Photomicrograph of radicular cyst showing arcading pattern (H&E, $\times 10$)

Fig. 6 Dilated enlarged follicle—single layer REE converting into a multilayer epithelial lining (H&E, $\times 10$)

Discussion

Over the years the opinion that radicular cysts arising from the deciduous teeth are rare has been commonly held. Sprawson [6] stated that radicular cysts arising from deciduous teeth are less often seen than those from permanent teeth, because of much shorter time that the deciduous teeth are present in the jaws. The information concerning the growth rate of radicular cysts is not well established but Livingston [7] has estimated the radicular cyst growth as 5 mm annually. Deciduous molar teeth affected by caries at a young age (4–5 years), resulting in loss of vitality if not removed, may remain in the jaw for an additional 5–6 years, a period believed to be sufficient to form a radicular cyst [8]. Benn and Altini [9] suggest that periapical inflammation from a non vital primary tooth may spread to involve the follicle of the permanent successor. The inflammatory exudate leads to the formation of a dentigerous cyst which are designated as inflammatory follicular cysts [10].

Treatment options include removal of the cyst in toto with extraction of the primary tooth or marsupialisation of the cyst to the oral cavity. Enucleation would risk the developing tooth bud whereas marsupialisation would allow for decompression of the cyst with preservation of the developing dentition. In addition, it would allow for root development and guided eruption of the tooth bud.

Overlapping cystic pathological changes question the success of a planned marsupialisation procedure, as an attempt to do so would probably only decompress the radicular cyst. The lingually concealed dilated follicle not evident on routine radiographic examination would, however, persist and may subsequently enlarge. With the tooth bud also being a part of non decompressed pathology, the chances of it erupting would remain highly unlikely. Moreover, the patient over a period of time could require an additional surgical procedure to remove the impacted tooth or to enucleate the cystic change.

It was interesting to note that both the cystic cavities were considerably large in size with a relatively distinct thick lining. Sound clinical judgement and decision making during the operative procedure would prevent the operating surgeon from leaving the other pathological tissue in situ, treating it as an enlarged follicle and giving a chance for the impacted developing tooth germ to erupt. On practical grounds the attached dilated follicular tissue would be worth considering a benign cystic change and be treated like one irrespective of the histopathological report.

Distinction should be made between widening of the follicular space that normally accompanies eruption, and early stages of cyst formation. Pericoronal radiolucencies that exceed 2.5 to 3.0 mm can be considered to be cystic [11]. According to Shear and Speight some unerupted teeth

have a slightly dilated follicle in the pre-eruptive phase. These do not signify a cyst, nor even necessarily a potential cyst unless the pericoronal width is at least 3–4 mm [12]. Radiographic features for early detection of developing pericoronal pathology include: follicular radiolucency greater than 2.5 cm in diameter, unerupted tooth with enlarged follicular space which is not in its normal eruption position, unerupted tooth with an enlarged follicular space where root formation is complete, and inferior border of the follicular space visible across the neck of the tooth [13].

Daley and Wysocki (1995) [13] have pointed out that it can be difficult to distinguish between a small dentigerous cyst and a large dental follicle despite the availability of both radiological and histological information. Their comparative study of 1662 dentigerous cysts and 824 dental follicles showed considerable overlap in age distribution and site predilection, and they concluded that distinguishing reliably between a small dentigerous cyst and a large dental follicle may only be resolved by identifying a cyst cavity at the surgical operation. Damante and Fluery (2001) [14] suggested that the first radiographic diagnosis for a pericoronal space enlargement, in most of the routine clinical cases, should be of 'inflammation of the follicle'. A differential diagnosis of 'dentigerous cyst' or 'paradental cyst' might be considered. The final differential diagnosis between a small dentigerous or a paradental cyst and a pericoronal follicle depends on clinical and/or surgical findings, such as the presence of bone cavitation and cystic content.

Radiographic images considered adequately sufficient for routine management of benign cystic pathologies would probably be insufficient to interpret the presence of an additional cystic change concealed by the larger one.

Presence of a well defined cystic cavity on the mesial side in this interesting situation does favour a cystic pathology over an enlarged follicle. Occurrence of a radicular cyst due to a decayed deciduous tooth is relatively uncommon, but its occurrence in conjunction with an additional cystic change is even rarer.

Certain questions do remain unanswered. Would the dilated enlarged follicle considering its size, if unexplored have continued to enlarge? Would the decompression of the radicular cyst with extraction of the primary tooth be sufficient to restrict the growth of the dilated follicle with a tooth bud displaced near the lingual border? Would it be worthwhile to attempt to preserve the tooth bud involved in this strange displaced presentation? These are all debatable issues.

To conclude, overlapping pathological cystic changes do spring up a surprise and can alter the treatment plan. What seemed to be a straightforward case of a cystic lesion with the primary dentition coincidentally had a concealed

secondary cystic pathological change within the same region. Sometimes there is much more hidden in the simplest of clinical situations than what the eyes and mind can comprehend. The rationale for marsupialisation is further questioned as the additional enlarged follicle would not have been explored if enucleation would not have been the primary treatment of choice in this particular case. This strange interesting presentation of two cystic pathologies overlapping each other with common zone of attachment is worth a mention as it has not been previously described in literature.

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