An Unusual Case of Radicular Cyst in a Primary Canine

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Abstract
Radicular cysts are common inflammatory odontogenic cysts that arise from epithelial cell rests of malassez due to periapical infection. They are usually encountered in association with permanent teeth. Radicular cyst arising from a primary tooth is exceedingly rare accounting for less than 1% of all radicular cysts [1]. Radicular cyst associated with primary anterior tooth is still rarer and very few cases have been reported in the literature till date [2,3]. As far as the authors are aware, this is the first report of radicular cyst associated with primary canine.

Keywords: radicular cyst, deciduous anterior tooth, maxillary canine, trauma, periapical cyst

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1. Introduction
Radicular cysts are common inflammatory odontogenic cysts that arise from epithelial cell rests of malassez due to periapical infection. They are usually encountered in association with permanent teeth. Radicular cyst arising from a primary tooth is exceedingly rare accounting for less than 1% of all radicular cysts [1]. Radicular cyst associated with primary anterior tooth is still rarer and very few cases have been reported in the literature till date [2,3]. As far as the authors are aware, this is the first report of radicular cyst associated with primary canine.

Radicular cysts are usually asymptomatic and are left unnoticed, until detected by routine radiography. The etiologic factor most commonly implicated is dental caries followed by trauma. However several authors [4,5,6,7,8] have reported radicular cyst associated with deciduous molar teeth following endodontic treatment using materials containing formocresol which usually elicits humoral and cell mediated response. Nagata et al [9] reported that 56% of the total radicular cysts associated with primary teeth have occurred following pulp therapy. The aim of this article is to report a rare occurrence of radicular cyst associated with a nonvital maxillary primary canine.

2. Case Report
A 9 year old girl reported to the department of Pedodontics with chief complaint of discoloured upper front tooth and painless swelling in the same region since 1 month. Her past medical history was insignificant and her past dental history revealed that she had undergone extraction of mandibular right second deciduous molar 2 years back which was followed by insertion of a space maintainer. History also revealed that the patient had a traumatic injury to the upper front teeth 3 years back, but she did not seek any dental treatment.

On extra oral examination, there was a diffuse, non-tender swelling, in the left infra orbital region measuring 3 x 3 cm. Intra orally, an irregular, smooth bony hard swelling was seen on the buccal aspect, extending from left primary canine to second molar (Figure 1).

Figure 1. Intra oral view of the cyst

The left primary canine was discoloured, suggestive of nonvitality and the patient experienced pain on vertical percussion, indicating the presence of a periapical pathology. Orthopantomograph showed well defined large periapical radiolucency with a thin sclerotic border in relation to the apex of primary canine (Figure 2).

The apical one third of the root was resorbed simulating physiologic resorption. The permanent canine was displaced in an oblique direction near the floor of the orbital cavity.
Figure 2. OPG showing the cystic lesion and displacement of permanent canine

Fine Needle Aspiration Cytology (FNAC) revealed straw coloured fluid. From the history, clinical, radiographic examination and FNAC report, a provisional diagnosis of radicular cyst was made. Routine blood investigations were carried out and the results were well within normal limits. Under local anaesthesia, primary canine was extracted (Figure 3) and it was found that the cyst wall was attached to the root surface.

Figure 3. Extraction of primary canine

Through the socket, the cyst was completely enucleated and the cyst lining was sent for histopathological examination. Primary closure was done after thorough debridement, irrigation and hemostasis. The patient was recalled after 7 days for suture removal. Nance palatal arch space maintainer was inserted after suture removal and the patient was followed up every 3 months. Post surgical phase was uneventful with no complications. Histopathological examination showed nonkeratinized stratified squamous epithelial lining with chronic inflammatory cells infiltration, discontinuous arcading pattern of the epithelial lining, hemosiderin pigmentation, and hyaline bodies. In the connective tissue, there were moderate amount of collagen bundles and blood vessels with infiltration of chronic inflammatory cells. These features were consistent with the clinical diagnosis of radicular cyst. A change in the position of the permanent canine was noticed 12 months after the surgery (Figure 4), suggestive of eruption of the tooth. Eruption of the permanent canine was evidenced 28 months after the surgery (Figure 5 and Figure 6).

Figure 4. Radiograph after 12 months of surgery

Figure 5. Eruption of canine

Figure 6. Continuation of root formation in permanent canine
The tooth has erupted in its normal position in the arch without any developmental abnormalities.

3. Discussion

A radicular cyst is the one which arises from the epithelial cell rests of molas present in the periodontal ligament as a result of inflammation. The inflammation occurs following pulpal death and the cyst arising from the cell rests is commonly seen at the apex of the involved tooth. Most of the radicular cysts are symptomless unless associated with concomitant infection and are usually diagnosed by routine radiography.

The occurrence of radicular cyst in primary dentition has been reported to be rare. Nevertheless, the occurrence of these cysts in deciduous anterior teeth is still lesser. A recent review reported a frequency of 13% in anterior maxilla and 3% in anterior mandible of the total radicular cysts in the deciduous dentition [9]. While Smith et al reported radicular cyst in maxillary primary incisor following trauma [2], Al-khayatt et al reported the same following replantation of avulsed primary tooth [3]. The present case is considered as the first report of radicular cyst in primary canine following trauma.

Numerous reasons have been stated for the low incidence of radicular cyst in deciduous dentition. These include the presence of deciduous teeth for a short period of time, easy drainage in deciduous teeth due to the presence of numerous accessory canals and negligence of radiolucencies in relation to deciduous teeth [6]. Further, the lesions tend to resolve on their own following extraction of associated tooth and hence are not submitted for histopathological examination [5].

Many authors have reported this pathology following endodontic treatment of the deciduous teeth [5,10,11,12]. Grundy, Adkins and Savage [13] reported a series of cases of radicular cysts associated with deciduous teeth that had been treated endodontically with materials containing formocresol. Formocresol combines with tissue proteins and forms a complex that is antigenic which in turn elicits a humoral and cell-mediated response.

However the reason for low occurrence of radicular cyst in primary anterior teeth is still not understood. Although pulpal necrosis following dental caries is the predominant etiologic factor in the development of a radicular cyst, this phenomenon rarely occurs in primary anterior teeth. Despite the high incidence of early childhood caries, leading to gross destruction of maxillary anterior teeth and pulpal death, the incidence of radicular cysts in such situations is significantly less. In contrast to this, few authors have reported radicular cyst in deciduous anterior teeth following traumatic injury [14,15]. Whereas many authors [16,17,18] have reported dentigerous cyst as a common sequelae following trauma to the primary predecessor, radicular cysts are comparatively rare. The reasons behind this controversy have not been clearly understood.

For a confirmatory diagnosis of a radicular cyst associated with a primary tooth, certain criteria have been put forth by Shear [1]. They include the presence of a non-vital deciduous tooth in close relation to the radiolucency or a record that such a relationship had been present in the past, the presence of a radicular cyst epithelial lining in the lesion during histopathologic examination and non-involvement of the crown of permanent successor in the cystic cavity.

This case report emphasises the need for frequent follow up after trauma to primary teeth. Lack of proper follow up can prevent early diagnosis of various complications arising as a result of trauma to primary teeth. When left unattended, the lesion may result in several complications including enamel hypoplasia, cessation of root development, eruption disturbances, displacement and damage to the permanent successor [15]. In the present case, on initial examination, displacement of the permanent canine was seen. Following early intervention, it was noted during the follow up visits that the canine was erupting normally in the arch. The absence of anomalies or hypoplastic defects in the erupted permanent canine in this case is noteworthy. Sequential radiographic follow up showed the continuation of the root development.

In children, healing of the osseous defects after a surgical procedure is always good as they have high propensity for bone regeneration [5]. In our case, this was evidenced through routine follow up and uneventful eruption of the permanent canine. To conclude, we present a rare case of radicular cyst associated with primary canine. Recognition of the possibilities of radicular cyst to occur in a primary anterior tooth as a response to trauma is indeed important for prevention of complications to the underlying permanent successor.

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