Localized Idiopathic Internal Resorption in the Primary Dentition.

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Case report: *We present the clinical and histological findings in a case of a six year old male patient who had unexplained sequential spontaneous abscesses associated with an unusual pattern of resorption, resulting in the loss of his non-carious second primary molars.* ***Conclusions:*** *Despite a wide differential diagnosis, the case represents unusual clinical and histological features of resorption in multiple primary teeth.*
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INTRODUCTION

We present a case with unusual clinical and histological features of internal and external resorption in multiple primary molar teeth. Idiopathic internal resorption is well described, and is usually seen in isolated teeth in the primary dentition. Many factors have been suggested in the rare cases of multiple affected teeth, including trauma and pulpal pathology,[1,2] but the etiology has not been established in most cases. One recent case suggested a link between internal resorption affecting multiple teeth and atopic dermatitis,[3] but this has not been confirmed. The present case is unusual inasmuch that only the second primary molars were affected, each presenting with the development of an abscess. A similar case has recently been described in a 3 year old Japanese boy, but affecting a wider range of teeth in the primary dentition and without symptoms.[4]

CASE REPORT

This patient first presented at 4 years of age with a fluctuant swelling adjacent to the upper left second primary molar tooth.

The swelling was drained and amoxicillin prescribed. In the absence of an obvious etiology such as dental caries or periodontal disease, the patient was referred to the local Dental Hospital.

The patient has asthma for which he is prescribed a salbutamol inhaler but otherwise is healthy and of average stature. On examination, an abscess was noted associated with the upper left second primary molar tooth. Clinically the crown form was normal with no discoloration. There was no evidence of dental caries or of generalised or localized periodontal disease. Radiographic examination confirmed the absence of caries, but demonstrated root resorption and a radiolucent area in the furcation. The patient had the upper left second primary molar tooth extracted and sent for histopathological examination.

This showed extensive resorption of the coronal dentin with osteodentin repair. The remainder of the coronal dentine had a prominent interglobular pattern. The morphology of the pulp chamber was relatively normal and the coronal pulp was vital. There was no evidence of caries but an abscess was present on the pulp chamber floor extending into the root canals. The roots had extensive areas of external resorption, but cellular cementum was present.

Over the subsequent two years the patient presented with abscesses related to the remaining second primary molar teeth. Sequential radiographs showed progressive linear calcification within the pulp chambers that followed the outline of the pulp, evident prior to the development of symptoms (Figure 1). The teeth were extracted and sent for histopathological examination which revealed extensive external and internal resorption and pulp/furcation abscesses similar to the first tooth with no evidence of dental caries (Figure 2). Osteoclasts were not identified. The resorption appeared to perforate into the periodontal ligament in the area of the bifurcation.

The calcification noted on the radiographs consisted of a "shell" of osteodentin within the pulp chamber. Clefts extended through the clinical crown to the amelodentinal

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junction and the dentin had a focally prominent interglobular mineralisation pattern. As in the previous tooth, external resorption was also present.

Differential diagnosis

The complex and varied histopathological features seen raised an extensive differential diagnosis including hypophosphatemia, hypophosphatasia, various forms of dentinogenesis imperfecta/dentin dysplasia and an unusual form of idiopathic internal resorption. The patient has been subjected to extensive medical examination which revealed no significant abnormalities.

The patient's serum biochemistry evaluation revealed: calcium 2.31mmol/l (normal range 2.20-2.70); phosphate 1.91mmol/l (normal range 0.90-1.80); alkaline phosphatase 197 (normal range 100-420).

Some histological features suggest hypophosphatemia (OMIM #193100), namely a large pulp chamber extending to the amelodentinal junction and prominent interglobular dentin. However, in the present case, the inflammation was primarily in the floor of the pulp chamber, rather than in the pulp horns. Additionally, the patient's serum biochemistry was within normal limits except for a slightly elevated phosphate, which does not support a diagnosis of hypophosphatemia. However, there are a number of other rare Vitamin D related syndromes in which the dental features have not been documented.[5] Similarly, whilst there are some features suggestive of hypophosphatasia (OMIM #146300), serum alkaline phosphatase is within normal levels and cementum formation has not been affected. Again, despite some suggestive clinical features, the histological features do not support a diagnosis of Dentinogenesis Imperfecta (DI) as the structure of the dentine is relatively normal.

Given the absence of known dental or systemic disease, we have classified this as idiopathic internal resorption. Previous cases reported have suggested indirect trauma to the tooth via a blow to the chin and bruxism may be possible causes.[3, 6] The patient reported no recent history of trauma to the mandible at any time and the observed pattern of the phenomenon in multiple teeth over 2 years suggests that this is unlikely. There were no convincing features suggestive of bruxism, such as prominent occlusal wear facets.

DISCUSSION

The extensive differential diagnosis raised by the radiological and histological features in this case reflects its complexity and demonstrates our lack of understanding of the physiological events in tooth resorption, and how these may relate to developmental disorders of tooth structure. We believe that these features are related to exaggerated physiological resorption, rather than a disorder of tooth development and as such these features represent an unusual presentation of idiopathic internal resorption. The reason why this process has been localized to the second primary molars is unclear as is the pathogenesis which has resulted in clefting and prominent interglobular dentin. The reparative deposition of calcified material in primary molars which have undergone extensive internal resorption has been reported,[7] but this is the first case in which the histopathological features have been described in detail.

This "osteodentin" response has also been reported in experimentally replanted incisors in cats and represents a manifestation of the healing process in the pulp.[8] A similar phenomenon has also been reported following pulpotomy in primary molar teeth.[9,10]

In this case, it appears likely that the teeth became symptomatic once the resoprtion extended into the periodontal ligament. This may have allowed communication with the oral cavity and the ingress of bacteria, provoking pulpitis and could account for the observed radiographic progression of the resorption prior to the development of symptoms. In view of our lack of understanding of the likely natural history of the internal resorption in this patient, we have protected the first permanent molars using Nobel gold

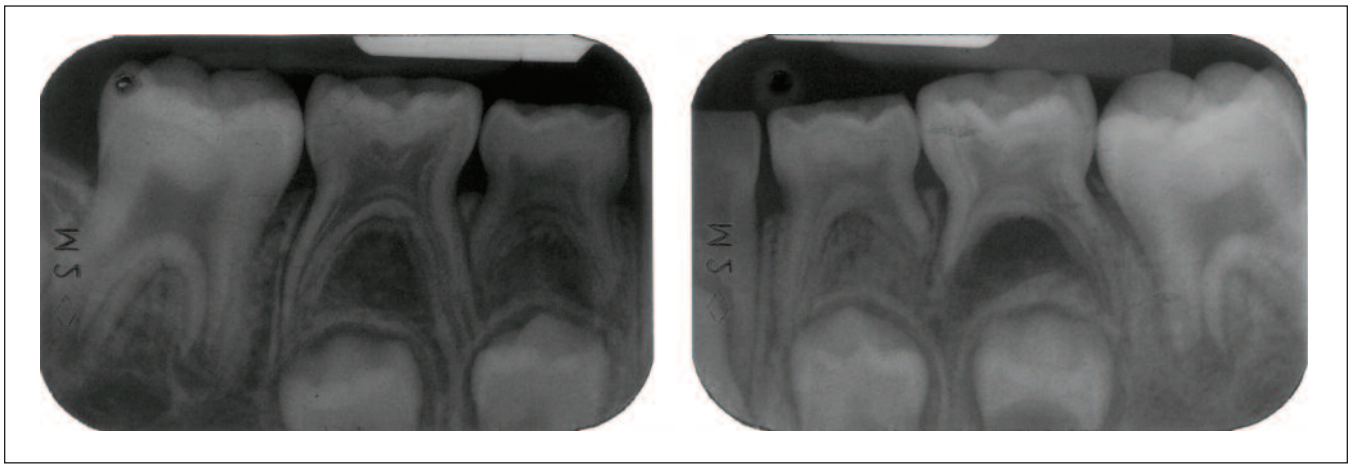

Figure 1. Intraoral radiographs of both mandibular posterior segments taken in August 2006. The radiopaque "shell" of osteodentin is evident in both lower second primary molar teeth, yet the patient did not develop symptoms for another 2 months.

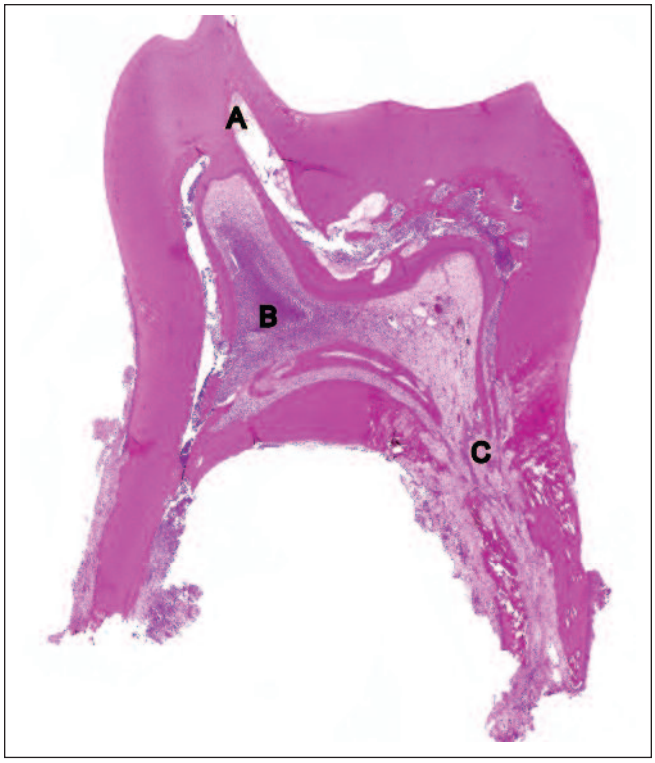

Figure 2. Photomicrograph of a hematoxylin and eosin stained decalcified section of the extracted lower left second primary molar, showing extensive clefting within the coronal dentin (A), extending towards the ADJ. There is also clear evidence of pulp inflammation (B) and root resorption (C). Similar features were seen in all affected teeth. (x10).

onlays to reduce the likelihood of bacterial ingress via cuspal clefts. Whilst we have assumed that the primary source of bacterial ingress has been from the periodontal ligament, dentine clefts extend to the ADJ and this is a further potential source of bacterial entry to the pulp. The patient has now been reviewed for 30 months with no further lesions evident. As a precaution, we have also advised that orthodontic tooth movement is avoided and the patient remains under long term follow-up.

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