

ORIGINAL PAPER

Delayed Exfoliation of Primary Teeth Due to Second Pathoses: Case Series Study

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ABSTRACT

The delayed exfoliation of primary teeth among children is a common and frequent dental problem whose most cited cause is misalignment of the crown of the successional permanent tooth with the root apex of the primary tooth. Treatment is often extraction of the over retained primary teeth and reassurance of the guardian that the normal tongue movements will push the misaligned permanent successional teeth into line in cases of mild misalignment or orthodontic treatment in severe cases of misalignment. However, meticulous history taking and careful clinical oral examination may yield an underlying pathosis as the three cases in this study illustrate. Underlying second pathoses may exert local and systemic effects on the normal exfoliative mechanism resulting in over retention of primary teeth despite proper alignment of the crown of the permanent successional tooth with the root apex of the primary tooth.

INTRODUCTION

The aetiology of over retention of primary teeth is often misalignment of the crown of the successional permanent tooth as it erupts relative to the root of the primary tooth. This explanation may be satisfactory to the 'concerned' parent or guardian but as the three cases in this study illustrate, there may be a second pathosis causing the over retention.

Case 1.

A seven-year old black female presented with over retention of 51, 52, 61, 62, 71 and 81 with ectopic eruption of the successional permanent teeth 11, 21, 22, 31 and 41. Poor oral hygiene was clinically evident as arrested and progressive carious lesions on all primary and permanent teeth except the permanent sixes. No periapical abscesses were evident radiographically nor draining sinuses and a diagnosis of rampant caries was made. Treatment rendered was oral health instruction, extraction under local anaesthesia of over retained primary teeth and restorations on the remainder. Interestingly, roots of the over retained 51, 52, 62 and 81 were intact showing no resorption as seen on the palatal and lingual aspects of 61 and 71 respectively.

Case 2.

A six year-old black female presented with a progressive swelling of the mandible of one-year duration. Extraoral examination showed mandibular prognathism and macrocheilia of the lower lip. Intraoral examination showed marked alveolar expansion of the anterior mandible on the labial aspect and overretention of primary teeth 71, 72, 81 and 82. The corresponding permanent successional teeth 31, 32, 41 and 42 had erupted on the lingual aspect of the deciduous teeth (Fig. 1.).

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Fig. 1. Case 2: Labial alveolar expansion and over retention of anterior incisors of mandible and secondary mandibular prognathism in 6-year-old black female.

Radiographic investigation showed a well demarcated unilocular radiolucent lesion of the anterior mandible. Laboratory investigations of serum found 48 U/L of alkaline phosphatase (normal 20-125 U/L), 1.03 mmol/L of calcium (normal 2.12-2.57mmol/L) and 9 g/L of albumin (normal 35-50g/L). A biopsy of the lesion under local anaesthesia was submitted for histological examination, which showed fibrous connective tissue stroma lined by 3 to 8 epithelial cell layers with occasional intraluminal heaping of the epithelial cells. Epithelial cells devoid of nuclei the so-called 'ghost cells' were evident in the superficial epithelial layers and the intraluminal aggregates. These features were interpreted as those of a calcifying odontogenic cyst. The case was subsequently referred to the oral and maxillofacial surgery clinic for further management.

Case 3.

A 12 year-old black female presented with a 5-year history of a painless right maxillary swelling that was noted on extraoral examination as facial asymmetry. Intraoral examination found a hard bony swelling limited to the buccal aspect of the right maxilla covered by normal coloured mucosa. Over retained primary teeth were present in the swollen right maxilla namely 52, 53, 54 and 55 (Fig.2) as well as labial to the successional permanent teeth in the mandible namely 73, 72, 71, 81, 82 and 83 (Fig.3). Laboratory investigations of

serum gave 393 U/L of alkaline phosphatase (normal 20-125 U/L), 2.49 mmol/L of calcium (normal 2.12-2.57 mmol/L) and 43 g/L of albumin (normal 35-50g/L). Radiographic examination showed a diffuse 'ground glass' radiopacity and expansion of the right maxilla. A subsequent biopsy under local anaesthesia of the alveolar bone of the swollen right maxilla was submitted for histological examination that showed irregular trabeculae of woven bone in fibrous connective tissue stroma; features that were diagnostic of monostotic fibrous dysplasia. Treatment rendered included extraction of the over retained primary teeth whose roots were intact and referral to an oral and maxillofacial surgery clinic for further management of the bony expansion and possible orthodontic exposure of the unerupted permanent teeth.



Fig. 2. Case 3: Firm bony expansion covered by normal coloured mucosa and over retention of primary teeth of right maxilla in 12-year-old black female.



Fig.3. Case 3. Over retained mandibular incisors in 12-year-old black female with monostotic fibrous dysplasia of the right maxilla.

DISCUSSION

Disturbed primary or deciduous tooth exfoliation can either be premature or delayed. This study illustrates the possible roles of second pathoses in the delayed exfoliation or over retention of primary teeth. The exact mechanism of primary tooth exfoliation could involve pressure resorption of the deciduous root invoked by the erupting successional tooth and or differentiation of monocytes of the periodontal ligament into odontoclasts. The odontoclasts then resorb the deciduous root in a similar manner to osteoclasts during bone remodeling or resorption with absence of an inflammatory response¹. The factor(s) that trigger this process remain unknown. The majority of carious deciduous teeth undergo normal exfoliation without restorations². It is postulated that a cumulative and quantitative effect of rampant caries may delay the start of the root resorptive process as case 1 shows with only 2 out of the 6 over retained teeth showing evidence of root resorption. Over retention of a deciduous tooth (84) due to the presence of a COC and an intraluminal adenomatoid odontogenic tumour has been reported³. Case 2 of this study documents a second case of COC involvement in over retention of four deciduous teeth 71, 72, 81 and 82. The COC of case 2 could have locally affected the exfoliative mechanism of deciduous teeth in the presence of reduced serum levels of calcium and albumin. The exact mechanism could have been the misalignment of successional teeth relative to the deciduous roots due to the cortical and alveolar cystic expansion, reduced serum levels of calcium and albumin or both. However, alveolar cystic expansion was the most likely explanation as it disturbed the integrity of the periodontal ligament (PDL) where the monocytes that differentiate into odontoclasts are located besides the physical misalignment.

Monostotic fibrous dysplasia (MFD) is a result of postnatal mutation of the guanine nucleotide-binding protein, alpha-stimulating activity polypeptide 1 (GNAS1) gene⁴. Case 3 had three times the normal alkaline phosphatase level while calcium and albumin were within normal ranges. Interestingly, the MFD of case 3 exerted both local and systemic effects during disruption of the mechanism of deciduous root resorption in the right maxilla whereas only the systemic effects could explain the over retention of the mandibular incisors. The bone trabeculae of fibrous dysplasia are considered to arise by metaplasia⁴ and in this case, the local effect could have been the metaplasia of alveolar bone of the right maxilla disturbing the

integrity of the PDL. It is theorized that systemic effects in both jaws but more so in the mandible could be the arrest of monocyte differentiation into odontoclasts, failure of the odontoclasts to attach to the deciduous roots' surface and lacunae formation, odontoclast failure to release the odontolytic enzymes into the lacunae or inactivation of released odontolytic enzymes due to the high serum alkaline phosphatase levels. Bone growth causes an age-dependent rise in alkaline phosphatase normal values particularly in children <2 yrs and adolescents. Thereafter, alkaline phosphatase activity declines reaching normal adult levels after a growth spurt during adolescence⁵. Three times the normal values of alkaline phosphatase in case 3 of the 12-year-old female cannot be adequately explained by early onset of puberty as the patient did not have physical signs of puberty at presentation. Chronic malnutrition reflected by a stunted growth pattern has been associated with delayed exfoliation of deciduous teeth⁶. The three children comprising this study showed no signs of chronic malnutrition and consequently malnutrition was ruled out in the differential diagnosis of the aetiology for the over retention of primary or deciduous teeth. In view of the unique aetiology for over retention of primary teeth and the female gender preponderance in this study, the number of cases does not support a factual female predominance of second pathoses delaying exfoliation of primary or deciduous teeth.

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