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CASE REPORT

An unusual combination of idiopathic generalized short-root anomaly associated with microdontia, taurodontia, multiple dens invaginatus, obliterated pulp chambers and infected cyst: a case report

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Generalized diminished root formation is a rare condition leading to early loss of teeth. This report describes an unusual case of generalized short roots associated with microdontia, taurodontism of posterior teeth, and multiple dens invaginatus along with short stature in a 20-year-old man, who had lost several teeth because of spontaneous exfoliation.
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Case report

A 20-year-old male presented with the chief complaint of missing lower anterior teeth and wanted replacement of the same. There was no history of trauma to the teeth. The patient had lost several teeth because of spontaneous exfoliation over a period of 3–4 years, resulting in aesthetic and phonetic problems. His medical history was unremarkable with no history of serious childhood illness or systemic abnormality. The family history did not reveal consanguineous marriage of his parents. According to his mother, the patient was born after an uneventful, full-term pregnancy with no exposure to radiation. Patient's parents and his siblings, an 18-year-old sister and a 14-year-old brother did not exhibit any similarity to the patient's dentition after thorough intraoral and radiographic examination.

On physical examination, the patient appeared relatively short with height (140 cm), weight (36 kg) and head circumference (45 cm), all in the lower limits of normal range for his age. The extraoral examination showed no abnormalities. Intraoral examination revealed 25 small, caries-free permanent tooth crowns of normal colour and shape (Fig. 1). Mandibular anteriors and maxillary left first premolar were absent. Maxillary right and mandibular left first premolars exhibited grade III mobility which were extracted later, while rest of the dentition showed grade-I mobility. His oral hygiene was good and mucosa appeared normal.

An orthopantomograph disclosed short, tapered roots with crown-to-root ratio of more than 1:1 in all the teeth but molars. There was an apical radiolucency associated with mandibular right first and second premolars, periapical tissue from which was curetted and sent for histopathological evaluation. The mandibular right and left first molars appeared to be taurodontic (Fig. 2). A full mouth radiographic survey revealed dens invaginatus in maxillary laterals, maxillary right first premolar and mandibular left first premolar. Pulp chambers of all the teeth except molars were partially obliterated, and that of maxillary centrals were radigraphically not appreciable (Fig. 3). Remnants of root tips were visible in the anterior mandible. Endocrinologic evaluation for growth hormone, thyroxin and cortisol were within the normal limits. Based on these findings, a provisional diagnosis of dentin dysplasia type I (radicular variety) was made.

Histopathological examination of curetted tissue from the area of apical radiolucency revealed features suggestive of an infected cyst. The extracted teeth exhibited a crown-to-root ratio of 1.4:1. Sagital section of mandibular left first premolar showed obliteration of pulp chamber with the presence of dens invaginatus (Fig. 4), whose ground section examination revealed normal enamel structure, normal tubular pattern of dentin with cementum covering the shortened irregular root (Fig. 5). Considering the histopathological findings, previous diagnosis of dentin dysplasia type I was ruled out and a final diagnosis of idiopathic generalized short-root anomaly was made.

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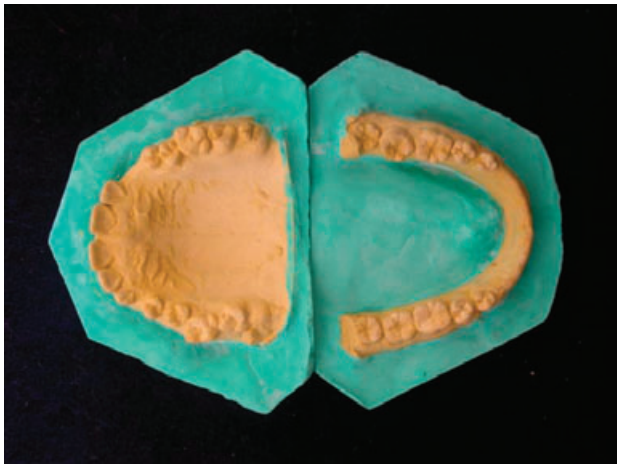

Figure 1 Casts showing generalized microdontia with missing teeth.

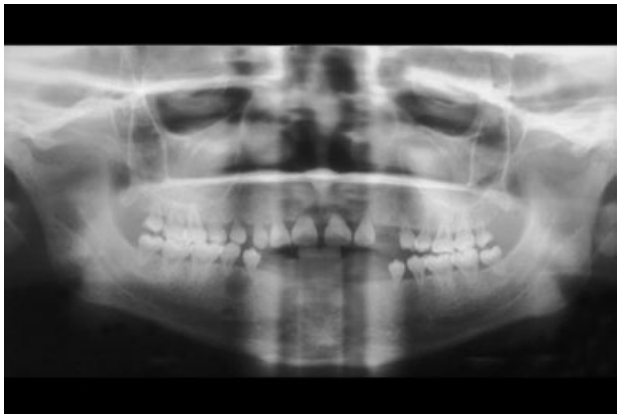

Figure 2 A panoramic radiograph showing generalized microdontia with short roots, taurodontia and periapical radiolucency.

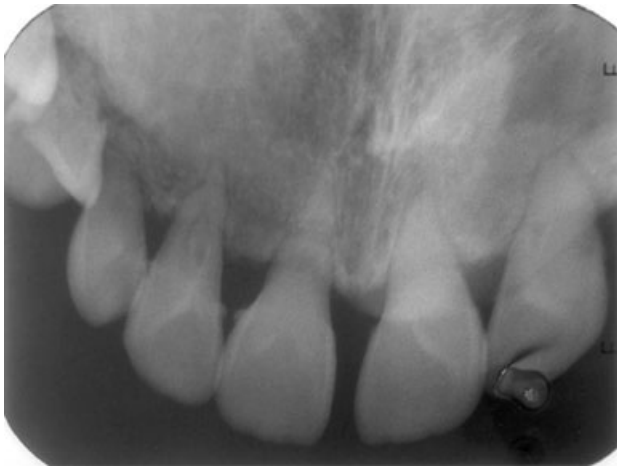

Figure 3 Intraoral periapical radiograph of maxillary anterior region showing short roots, obliteration of pulp chambers and dens invaginatus.

Comments

Short-root anomaly, occurring mostly in permanent maxillary incisors has been described as teeth having developmentally very short roots with a crown-to-root ratio of more than 1:1.6 (1). Short roots may also be observed in disorders like dentin dysplasia type 1 (2), scleroderma, Stevens–Johnson syndrome, Laurence–Moon–Bardet–Biedl syndrome, thalassemia, and in some short-stature syndromes such as, Down syndrome, Aarskog syndrome, dwarfism of Seckel and Rothmund–Thomson syndrome (3).

Idiopathic generalized short-root anomaly is extremely rare. MEDLINE search in the English literature

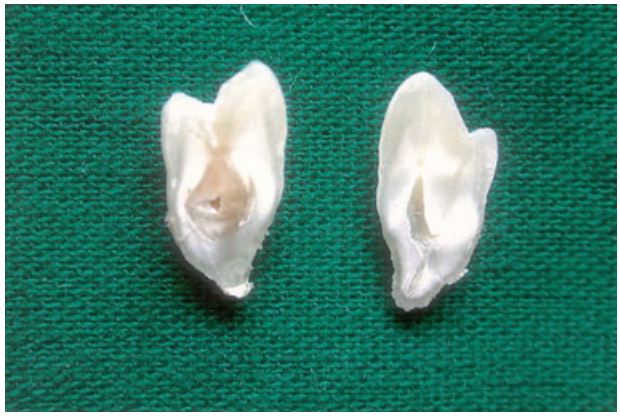

Figure 4 Sagital section of an extracted premolar showing obliterated pulp chamber with the presence of dens invaginatus.

Figure 5 Photomicrograph of ground section of extracted tooth showing dens invaginatus, normal enamel and normal tubular dentin.

for generalized short-root anomaly revealed seven reported cases, of which, two-exhibited familial tendency with autosomal dominant pattern of inheritance (4, 5), and the rest were sporadic cases (6–10). In our case none of the family members were similarly affected.

Generalized short-root formation, accompanied by taurodontia, multiple dens invaginatus (9), microcephalic dwarfism (7, 8, 10), as well as a prominent feature of dentin dysplasia type 1, has been described (2). Periapical radiolucency caused by external root resorption has been observed (8), but none of the published cases manifests periapical cyst formation as seen in our case.

Our case exhibits a peculiar combination of all the traits associated with short-root anomaly including generalized microdontia, taurodontism, multiple dens invaginatus, periapical radiolucency, obliteration of pulp chambers and increased tooth mobility leading to spontaneous exfoliation in a single patient. To the best of our knowledge, such unusual combination of multiple dental anomaly has not been reported so for in the literature. We also highlight the need for histopathological investigation in the form of ground and decalcified sections of teeth to negate the possibility of dentin dysplasia type 1 in patients with short roots.

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