

CASE REPORT

An unusual case of non-syndromic occurrence of multiple dental anomalies

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ABSTRACT

Dental anomalies have been known to occur in humans due to a variety of genetic and environmental factors. Combinations of dental anomalies are known to be associated with specific syndromes. A few cases of multiple dental anomalies have been reported in patients with no generalized abnormalities. This case report describes an unusual occurrence of a combination of dental anomalies in an apparently normal healthy 12-year-old female patient. The dental anomalies in this patient were multiple dens invaginatus, generalized enamel hypoplasia, generalized microdontia, root resorption and multiple periapical lesions, shovel shaped incisors, cup shaped premolars, taurodontism, hypodontia and supernumerary teeth.

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Dental anomalies have been known to occur in man due to a variety of genetic and environmental factors.¹ The simultaneous presence of multiple dental anomalies has been previously reported, especially in patients with chromosomal alterations and associated with multi-systemic alterations. A combination of dental anomalies has also been reported in patients with no generalized abnormalities or disease. While some cases of multiple dental anomalies within families have been reported, some may occur in individuals with no positive family history.²⁻⁴ Desai et al⁵ described an unusual case of generalized short roots associated with microdontia, taurodontism of posterior teeth, multiple dens invaginatus, spontaneous exfoliation of teeth due to root resorption, along with short stature. Casamassimo et al.⁶ reported an unusual triad of microdontia, taurodontia and dens invaginatus. The patient had normal appearance and no other illness. The pedigree was compatible with X-linked recessive inheritance. This case report describes an unusual occurrence of a combination of dental anomalies in an apparently normal healthy individual.

CASE REPORT

A 12-year-old female presented with a complaint of pain and swelling in the lower anterior region since three days. She had similar episodes of swelling with pain for the past two years. She had visited a dentist for the same for which she had been prescribed antibiotics. Sensitivity to cold food was perceived on all her teeth. There was no history of bleeding gums. Medical history did not reveal any history of serious

childhood illness or systemic abnormality. She was born of an uneventful full term pregnancy to consanguineous parents. Intra-oral examination revealed the presence of following teeth [Figures 1 and 2]:

7	6	4	3	2	1		1	2	3	4	6	7		
7	6	5	4	3	2	1		1	2	C	3	4	5	6

Various dental anomalies found clinical examination were:

- Supernumerary tooth resembling a premolar between 34 and 35.
- Microdontia with spaces in between the teeth.
- All teeth were hypoplastic with diffuse pitting on the labial/buccal surface and yellowish discoloration.
- The incisors and canines of both maxillary and mandibular arch were shovel shaped. The incisal edges were irregular.
- The molars and premolars were cup shaped with rosette like appearance in the occlusal view. The occlusal table was reduced in area due to small tooth size and was essentially flat because of the absence of cusp morphology [Figure 3].
- A diffuse, soft fluctuant swelling was seen on the labial gingiva of the lower incisors [Figure 2]. The lower incisors were tender on percussion and showed grade II mobility.

Various radiographic (Orthopantomogram and intraoral periapical radiographs) features are as follows [Figures 4 and 5]:

- Congenitally missing 25 and all third molars.
- Unerupted 15.
- Premolar-like supernumerary tooth between 34 and 35,

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with an abnormally long root.
Erupting premolar-like supernumerary teeth in proximity to 46.
Dens invaginatus of all the teeth except molars.
Radiodensity of enamel of anterior teeth was similar to that of dentin.
Diffuse periapical radiolucency in relation to 31,41; 11, 21; and 34, 35, supernumerary tooth, 36.

Taurodontism in 45.
Root resorption of 16, 36, 44 and 46.
All molars were single rooted.

As a number of dental anomalies were seen on examination, a systemic cause was suspected. The patient was referred to the pediatrician for a thorough medical examination and assessment of growth. She was found to be in normal

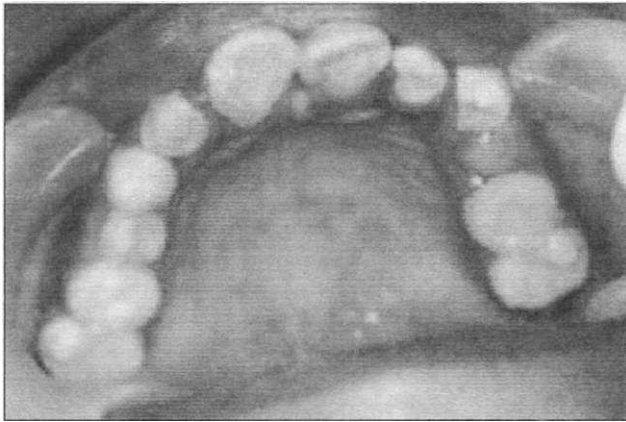


Figure 1: Intra oral view of the upper arch



Figure 2: Intra oral view of the lower arch showing the presence of intra oral swelling

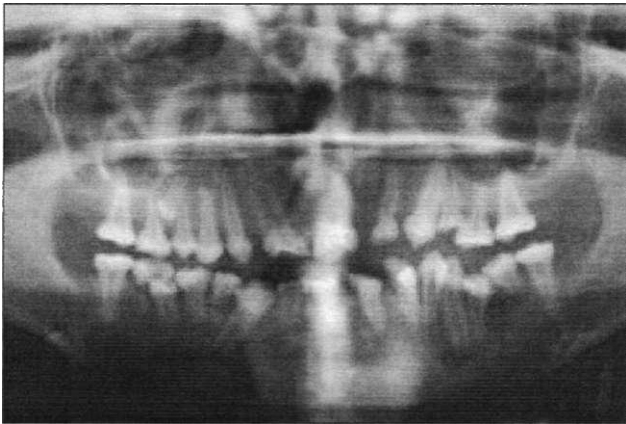


Figure 4: Orthopantomograph of the patient

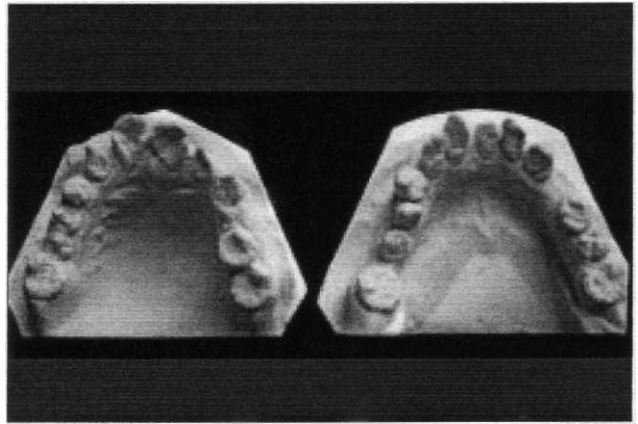


Figure 3: Diagnostic casts showing the morphology of the teeth

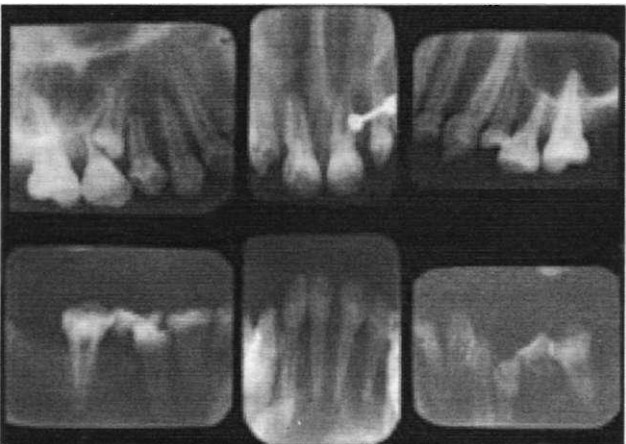


Figure 5: Intra oral periapical radiographs of the patient

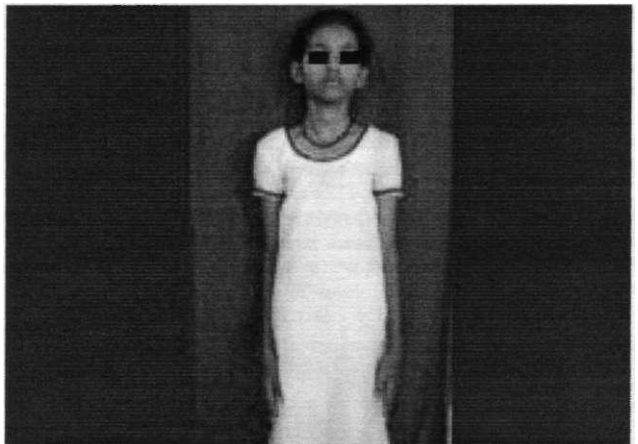


Figure 6: Frontal view of the patient

percentile of growth and no other abnormal findings were noted [Figure 6]. Her blood profile was normal. Serum calcium, phosphorous and alkaline phosphatase levels were also normal. Endocrinological evaluation showed that hormone levels were within normal limits. None of her family members had similar findings. According to history given by the parent, similar abnormalities were not noted in her primary dentition. As no systemic abnormalities were

dental anomalies was made.

It was decided to render corrective and preventive dental treatment for her dental problems. The intra oral abscess was drained and root canal treatment was completed on 31 and 41. The lesion responded well to conventional root canal therapy. While 11, 21 and 34 also responded well to conventional root canal therapy, 73 and 75, 36 and 46 were extracted due to extreme mobility and root resorption. Ground section and H and E sections of these teeth did not show any abnormality in the structure of enamel, dentin or cementum. During follow-up appointments (every three months over a period of 2 years), oral prophylaxis was done and oral hygiene instructions were given. The large radiolucency in relation to 36 disappeared and normal bone healing was seen following extraction of 36. The patient has been asked to continue the follow-up visits.

DISCUSSION

Dental anomalies in this patient were multiple dens invaginatus, generalized enamel hypoplasia, generalized microdontia, root resorption and multiple periapical lesions, shovel shaped incisors, cup shaped premolars, taurodontism, hypodontia and supernumerary teeth. According to our knowledge, no scientific literature is available on a similar pattern of dental anomalies in a single individual. The differential diagnosis included hypoparathyroidism, psuedohypoparathyroidism, vitamin D resistant rickets, hypophosphatasia, oculodontoosseus dysplasia, dystrophic epidermolysis bullosa, trichodontoosseous syndrome, tuberous sclerosis, dwarfism, Russell silver syndrome, Seckel syndrome, William's syndrome and amelogenesis imperfecta.¹⁶⁻¹⁸ As the patient did not show any abnormal systemic manifestations, all the syndromes associated with the dental anomalies were ruled out. Multiple peri-apical lesions with shovel shaped incisors are known to occur in dentin dysplasia Type I (radicular variety).¹⁹ However, the absence of short roots anomaly and the histopathological evaluation ruled out the diagnosis of dentin dysplasia. Taurodontism, short roots and external resorption have been described in patients with small head and short stature.¹⁰ In the present case, the physical growth was within normal limits and no short root anomaly was seen.

In teeth with dens invaginatus, the invagination is a pathway for irritants and microorganisms. Constant irritation results in pulp necrosis and apical abscess, which was seen

in the current case. This can be prevented by sealing the invagination with a restorative material and maintaining a good oral hygiene.¹⁰ In the above case, patient's oral hygiene was poor due to hypoplastic teeth with rough pitted enamel and many teeth had dens invaginatus which explains the multiple periapical lesions.

The combination of dental anomalies, as seen in this case,

etiology giving rise to different phenotypic manifestations. The constellation of dental findings seen in this case is certainly rare and differs from previously reported cases. The case is also sporadic, with no positive family history. Mutations in developmental regulatory genes are known to cause a variety of dental defects.²¹ The wide variation in clinical manifestations in cases of non-syndromic occurrence of multiple dental anomalies remains intriguing and is an area for further research. For the clinician, the difficulty lies in diagnosis, as a number of syndromes and pathologies need to be ruled out, due to large constellation of dental findings.

REFERENCES

1. Ezoddini AF, Sheikha MH, Ahmadi H. Prevalence of dental developmental anomalies: A radiographic study. *Community Dent Health* 2007;24:140-4.
2. Desai RS, Vanaki SS, Puranik RS, Rashmi CS, Nidawani P. 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, *J Oral Pathol Med* 2006;35:407-9.
3. Acs GA, Pokala P. Constellation of dental anomalies in a chromosomal deletion syndrome (7q32): Case report. *Pediatr Dent* 1994;16:306-9.
4. Ekman-Westborg B, Julin P. Multiple dental anomalies in dental morphology: Macrodontia, multituberculism, central cusps and pulp invaginations. *Oral Surg Oral Med Oral Pathol* 1974;38:217-22.
5. Casamassimo PS, Nowak AJ, Ettinger RL, Schlenker DJ. An unusual triad: Microdontia, taurodontia, and dens invaginatus. *Oral Surg Oral Med Oral Pathol* 1978;45:107-12.
6. Tiku A, Damle SG, Nadkarni UM, Kalaskar RR. Hypertaurodontism in molars and premolars: Management of two cases. *J Indian Soc Pedo PrevDent* 2003;21:131-4.
7. Tsai SJ, King NM. A catalogue of anomalies and traits of the permanent dentition of southern Chinese. *J Clin Pediatr Dent* 1998;22:185-94.
8. Hall RK. Abnormalities of tooth structure and appearance In: Hall RK, editor. *Pediatric Orofacial Medicine and Pathology*. London: Chapman and Hall Medical; 1994. p.184-224.
9. Witkop CJ. Amelogenesis Imperfecta, dentinogenesis imperfecta and dentin dysplasia revisited: Problems in classification, *J Oral Pathol* 1988;17:547-33.
10. Gardner DC, Cirgis SS. Taurodontism, short roots and external resorption associated with short stature and a small head. *Oral Surg Oral Med Oral Pathol* 1977;44:271-3.
11. Galindo-Moreno PA, Parra-Vazquez Mj, Sanchez-Fernandez E, Avila-Ortiz GA. Maxillary cyst associated with an invaginated tooth: A case report and literature review. *Quintessence Int* 2003;34:509-14.
12. Thesleff I. Genetic basis of development and dental defects. *Acta Odontol Scand* 2000;58:191-4.

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