#### Journal of Dental Research, Dental Clinics, Dental Prospects

# Case Report

# **Regional Odontodysplasia: A Case Report**

Saadettin Dagistan<sup>1</sup> • Ozkan Miloglu<sup>1</sup>\* • Mustafa Goregen<sup>2</sup> • Abubekir Harorli<sup>3</sup>

<sup>1</sup>Assistant Professor, Department of Oral Diagnosis and Radiology, Faculty of Dentistry, Ataturk University, Erzurum, Turkey <sup>2</sup>DDS, Department of Oral Diagnosis and Radiology, Faculty of Dentistry, Ataturk University, Erzurum, Turkey <sup>3</sup>Professor, Department of Oral Diagnosis and Radiology, Faculty of Dentistry, Ataturk University, Erzurum, Turkey \*Corresponding Author; E-mail: omiloglu@hotmail.com

Received: 16 September 2009; Accepted: 11 November 2009 J Dent Res Dent Clin Dent Prospect 2009; 3(4):141-144 This article is available from: http://dentistry.tbzmed.ac.ir/joddd

© 2009 The Authors; Tabriz University of Medical Sciences

This is an Open Access article distributed under the terms of the Creative Commons Attribution License (http://creativecommons.org/licenses/by/3.0), which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.

# Abstract

Regional odontodysplasia (RO) is a rare developmental dental anomaly with an unknown etiology. It is more often seen in girls than boys. Treatment of RO depends on the individual case. The aims of treatment should include aiding mastication and speech, improving aesthetics, reducing the psychological impact of the anomaly, allowing normal jaw growth and development, and if possible protection of any erupted teeth which are affected. We present a rare case of RO together with the treatment modality undertaken.

Key words: Odontodysplasia, odontogenesis imperfecta, regional odontodysplasia, treatment.

# Introduction

**R**egional odontodysplasia (RO) is a rare developmental dental anomaly. This disorder was first reported by McCall and Wald in 1947.<sup>1</sup> But the term of odontodysplasia was used for the first time by Zegarelli et al in 1963.<sup>1,2</sup> Because the disorder affects a certain segment of the jaws, the definition of RO is commonly used.<sup>3</sup> This disorder is additionally defined as odontodysplasia,<sup>2,4</sup> odontogenesis imperfecta,<sup>2,4-6</sup> unilateral dental malformation and ghost teeth.<sup>2,4-7</sup>

Its etiology is not exactly known,<sup>1-3,5,8</sup> but possible causes include local trauma,<sup>2,3,8</sup> infection,<sup>2</sup> local ischemia,<sup>3,5</sup> local vascular defect,<sup>2,3,8</sup> rhinocompatibility,<sup>2,8</sup> exposure to radiation,<sup>2</sup> nerve damage,<sup>2,5</sup> high fever,<sup>2</sup> local infection,<sup>3</sup> malnutrition and metabolism,<sup>2,3</sup> heredity, local somatic mutation and epidermal nevus syndrome.<sup>2,3,5,7,8</sup> In addition, some medications have been reported as possible causes <sup>5</sup> and latent viral syndromes in tooth germs have been considered as etiologic agents.<sup>3,5,8</sup>

Odontodysplasia develops as a result of local stagnation in tooth development. The disease involves mesodermal and ectodermal layers of teeth.<sup>9,10</sup> The impairment in the teeth affects both dentin and enamel.<sup>11</sup> The layers of dentin and enamel are tiny, and pulp chambers are wide.<sup>11</sup>

Hypocalcification, hypoplasia and discoloration are detected in the teeth affected by odontodysplasia.<sup>6,12,13</sup> The eruption of affected teeth is delayed.<sup>6,10</sup> Eruption may not occur in severe cases.<sup>6</sup>

We present a rare case of the developmental anomaly called RO or "ghost teeth" in a 6-year-old boy.

# Case report

Our patient was a six-year-old boy and was the 8th child of the family. He attended our clinic complaining of delayed eruption of teeth in the left mandible.

#### 142 Dagistan et al.

All the deliveries of the mother (a total of nine deliveries) were normal. The mother had urinary infection during pregnancy. The mother had been smoking a packet of cigarettes every two days, including the pregnancy period. When the patient was one year old, he had had convulsions and had been prescribed Depakin (SANOFI-SYNTHELABO) for four years, and then the medicine had been withdrawn. He had not been taking any medications for a year when he attended our clinic. Medical examinations did not reveal any systemic problems and he was in a generally good condition. He had speech disorder and his facial skin was normal.

In clinical examination, although the upper half of the face was normal, there was asymmetry in the lower half (Figure 1). There were no abnormal findings in the nails and eyes. In the maxillary arch, there were partially erupted left primary lateral incisors, canines, and first and second molars and right primary second molars in addition to the right first permanent molars. The other permanent incisors, canines, premolars and molars were unerupted. In the mandibular arch, only the right first and second primary molars and the right incisor were present; these teeth were clinically normal. The lower right primary lateral incisor and canine, left primary central and lateral incisors, canine, first and second molars were absent (Figure 2a and 2b)

In panoramic radiographic examination (Figure 3a) and periapical radiographs (Figure 3b), the maxillary dentition and right region of mandibular dentition were normal. The teeth in the left region of mandibular dentition had thin radiopaque contours with no distinction between enamel and dentin, and wide pulp chambers, giving a 'ghost-like' appearance. The crowns of the affected teeth were surrounded by large radiolucent areas, probably representing enlarged dental follicles. Diagnosis of RO was made on the basis of the clinical and radiographic findings.

Figure 1. Frontal view of the patient. JODDD, Vol. 3, No. 4 Autumn 2009

The decayed teeth were restored. Maxillary left primary first molar tooth and maxillary right primary second molar received full crowns (Figure 4), and partial dentures were fabricated for upper and lower jaw (Figure 5). Oral hygiene instructions and dietary



Figure 2. Intraoral images of maxillary (a) and mandibular (b) arches of the patient.



Figure 3. Panoramic (a) and periapical (b) radiographs of the patient.



Figure 4. Intraoral image of the patient after delivery of full crowns.



Figure 5. Intraoral image of the patient after fabrication of partial dentures for upper and lower jaws.

recommendations were given. The patient was called in at certain intervals (once every 6 months).

# Discussion

The etiology of RO is not known exactly. However, it has been reported that some medications administered for systemic and local reasons might play a role in the etiology of this disease.<sup>1-3,5,8</sup> Although the precise etiological factor in this case is unknown, the pattern of involvement suggests that the unknown damaging factor has been persistent, presumably starting before birth and continuing to act right through the period of development of the lower permanent incisors and canines.

Odontodysplasia is often seen in both permanent and primary dentitions.<sup>6,7,12,13</sup> Generally, the disorder is localized to one arch and maxilla is involved twice as often as mandible.<sup>14</sup> However, in this case, the teeth demonstrating odontodysplasia were in the lower arch. In both maxilla and mandible, central and lateral incisors, and canines are the teeth most commonly affected.<sup>4,6,10,12,13,15</sup> In the case we presented in addition to incisors, canines and molars were involved, which makes the case more interesting.

Teeth with RO sometimes may not be distinguished from the ones with dentinogenesis imperfecta. There is a generally familial involvement history in dentinogenesis imperfecta. In addition, there is remarkable hypoplasia in enamel in regional RO, which is not the case in dentinogenesis imperfecta.<sup>13</sup> Although conditions such as dentinal dysplasia and amelogenesis imperfecta show some similarities to RO, these conditions affect the entire dentition without segmental involvement. When the affected teeth erupt, RO can be mistaken for dental caries; therefore, the teeth might be extracted without being submitted for histological examination.<sup>14</sup> Many cases of RO are also misdiagnosed as malformed teeth or odontomas.<sup>16</sup>

There are various ideas about the treatment of odontodysplasia. While a number of clinicians suggest that

the teeth affected should be immediately extracted and dentures should be fabricated, some other clinicians recommend restorative procedures in an attempt to protect the affected teeth as soon as possible.<sup>1</sup> Those who suggest the extraction of the teeth claim that the teeth affected become, most of the time, abscessed, and painful, and defective enamel and dentin cannot be protected against bacterial invasion; as a result, the risk of developing necrosis and facial cellulitis may be high in near future.<sup>3</sup> Those who suggest restorative procedures recommend that the involved teeth should be restored and preserved in the mouth. Teeth with delayed eruption should be preserved in the mouth during skeletal growth. Severely damaged permanent teeth with pulpal involvement can be extracted and dentures can be fabricated.<sup>13</sup>

We believe the affected teeth in these patients should be restored as much as possible and retained in the mouth. However, severely damaged permanent teeth with pulpal involvement should be extracted and dentures fabricated, if necessary, to provide function and esthetics for the patients.

#### Acknowledgement

We would like to express our sincere gratitude to Lecturer Ali Çağlar Güllüce for his support on proof reading of our article.

# References

- 1. Cahuana A, González Y, Palma C. Clinical management of regional odontodysplasia. *Pediatr Dent* 2005;27:34-9.
- Pandis N, Palido C, Bell WH. Regional odontodysplasia: a case associated with asymmetric maxillary and mandibular development. *Oral Surg Oral Med Oral Pathol* 1991;72:492-6.
- Rosa MC, Marcelino GA, Belchior RS, Souza AP, Parizotto SC. Regional odontodysplasia: report of case. J Clin Pediatr Dent 2006;30:333-6.
- 4. Shafer WG, Hine MK, Levy BM, Tomich CE. *A Textbook of Oral Pathology*, 4th ed. WB Saunders Company: Philadelphia; 1983:63-4.
- Courson F, Bdeoui F, Danan M, Degrange M, Gogly B. Regional odontodysplasia: Expression of matrix metalloproteinases and their natural inhibitors. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2003;95:60-6.
- 6. Brooks SL, Kathryn AA. Guidelines for prescribing dental radiographs. In: Goaz PW, White SC. *Oral Radiology: Principles and Interpretation*, 5th ed. St. Louis: Mosby; 2004.
- Abnormalities of teeth. In: Neville BW, Damm DD, Allen CM, Bouquot JE, eds. Oral & Maxillofacial Pathology, Vol. 2. WB Saunders Co: Philadelphia; 2002:70.
- Dahllof G, Lindskog S, Theorell K, Ussisoo R. Concomitant regional odontodysplasia and hydrocephalus. *Oral Surg Oral Med Oral Pathol* 1987;63:354-7.
- 9. Raez AG. Unilateral regional odontodysplasia with ipsilateral mandibular malformation. *Oral Surg Oral Med Oral Pathol* 1990;69:720-2.

#### 144 Dagistan et al.

- 10. Laskaris G. Color Atlas of Oral Diseases in Children and Adolescents. Thieme: Stuttgart; 2000:32-3.
- Cawson RA, Binnie WH, Barrett AW, Wright JM. Oral Disease: Clinical and Pathological Correlations, 4th ed. CV Mosby Co: Edinburgh; 2001:22-3.
- 12. Fanibunda KB, Savers JV. Odontodysplasia, gingival manifestation, and accompanying abnormalities. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1996;81:84-8.
- 13. White SC, Pharoah MJ. Oral radiology: Principles and Interpretation, 4th ed. CV Mosby Co: St. Louis; 2000:349.
- Volpato L, Botelho G, Casela L, Borges A, Silva K. Regional odontodysplasia: report of a case in the mandible crossing the midline. J Contemp Dent Pract 2008;9:142-8.
- Gomes MP, Modesto A, Cardoso AS, Hespanhol W. Regional odontodysplasia: report of a case involving two separate affected areas. ASDC J Dent Child 1999;66:203-7.
- Ferguson F, Schlissel E, Kucine A, Alexander S, DeSantis A, Hendricks R, et al. Long-term management of a child with regional odontodysplasia: a case report. *Pediatr Dent* 2009; 31: 346-9.

#### Erratum

The article entitled "Assessment of the etiologic factors of gingival recession in a group of patients in Northwest Iran" which appeared in *J Dent Res Dent Clin Dent Prospect* 2009; 3(3):90-93 did not list the fourth author's name. The journal regrets this error. The correct list of authors of this article is as follows: Ardeshir Lafzi, Nader Abolfazli, Amir Eskandari, and Mehrnoosh Sadighi. Dr. Mehrnoosh Sadighi is a post-graduate student at the Department of Periodontics, Faculty of Dentistry, Tabriz University of Medical Sciences.