



# An alternative restorative method for regional odontodysplasia: case report

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### Abstract

A 5-year-old Caucasian male presented with early loss of multiple deciduous teeth. All the characteristics were consistent with the diagnosis of regional odontodysplasia (ROD). Significant initial findings included premature loss of multiple primary mandibular teeth and some malformed permanent teeth. The affected teeth showed hypoplastic enamel and dentin, short roots, and wide pulp chambers, and were localized in the mandibular dentition. Treatment objectives for this patient were to provide improved esthetics, restored chewing function, and space maintenance by the construction of a temporary prosthetic restoration. However, with limited tooth support and an unusual occlusal pattern, it is difficult to obtain satisfactory retention and esthetics with traditional prosthetic techniques. In this article we introduce an alternative method for fabricating a custom removable denture and discuss the prognosis of the malformed permanent dentition and further treatment plan. (*Pediatr Dent* 19:421–24, 1997)

Regional odontodysplasia (ROD) is a relatively rare dental anomaly. The clinical manifestations of odontodysplasia are:<sup>1</sup>

- Hypoplasia and hypocalcification of the enamel and dentin
- Localized occurrence within a particular segment in either or both dentitions
- Discolored, small, and distorted affected teeth with various surface markings (e.g., irregular contour, shallow furrows, pitting, and grooves)
- Delayed or failed eruption due to arrested root formation.

The radiographic features have consistently demonstrated thin and defective layers of enamel and dentin, resulting in a faint, fuzzy outline, creating a ghost-like appearance. The pulp chambers and canals are enlarged and the roots appear short and stubby with open apices. True and false denticles may be seen in the pulp of affected teeth as well as adjacent, clinically normal teeth.<sup>1</sup>

The first report of this condition was published by McCall and Wald<sup>2</sup> under the title of *Arrested Tooth De-*

*velopment*, and reported only radiographic findings. The first comprehensive description of this condition was by Hitchin,<sup>3</sup> who published a description of unerupted primary teeth. The term odontodysplasia was introduced by Zegarelli, et al.<sup>4</sup> Numerous case reports and literature reviews have been published since then. An article by Crawford and Aldred provides a relatively complete bibliography of the condition.<sup>1</sup>

Published cases suggest ROD occurs more frequently in females, with no racial predilection. The condition can affect both primary and permanent dentitions and can occur in the maxilla, the mandible, or both. The maxilla is involved twice as often. Although the incisors and canines are most often involved, any tooth may be affected. When primary teeth are involved, the permanent successors are also usually affected.<sup>1</sup> The condition tends to affect several adjacent teeth within a particular quadrant. Lustmann<sup>5</sup> found that affected teeth were located on both sides of the midline in only 11 of the 51 cases reported. ROD is usually seen in consecutive teeth, with no normal teeth interposed, although occasional skipped or minimally affected teeth do occur in the permanent dentition. Permanent first molars are affected only if the adjacent primary molar exhibits ROD.<sup>6</sup>

Multiple etiologies have been proposed, with defective vascular function the most widely accepted. The association of vascular nevi in the facial skin overlying areas where ROD occurs suggests that a local vascular defect may be involved in the pathogenesis of ROD. These nevi are pale pink in color and disappear gradually with age.<sup>7</sup>

Other conditions which share some characteristics with ROD include amelogenesis imperfecta, dentinogenesis imperfecta, dentinal dysplasia types I and II, shell teeth, rickets, and hypophosphatasia. However, it should be noted that, unlike ROD, all these conditions affect the entire dentition rather than a localized area. Turner's teeth should be considered if only one or two permanent teeth are affected.<sup>1</sup>

Though many cases of ROD have been reported in the literature, few have addressed treatment of this condition. Usually, cases were treated by the extraction of the affected teeth and restoration with a temporary

partial denture. Prosthetic management of multiple missing primary teeth is difficult because of the dynamic condition of the primary dentition (e.g., growth, exfoliation, and transition to permanent dentition). Although dental implants are advocated for restoration of adult patients, they have not been proven suitable for growing children. Therefore, a removable denture is the best available treatment choice to establish chewing function, normal vertical dimension, space preservation, and improved esthetics. This report introduces an alternative method of denture fabrication for patients with multiple missing primary teeth, abnormal tooth morphology, and unusual occlusal relationship. Rather than extracting affected teeth, preventive root canal treatment and restoration with stainless-steel crowns were done to preserve these teeth by protecting them from infection.

## Case report

### History and chief complaint

A 5-year-old Caucasian male was referred from a private dentist to the pediatric dental clinic of the University of Alabama at Birmingham with complaints of premature loss of multiple primary teeth and malformation of permanent teeth. The patient's private dentist reported that several hypoplastic primary teeth, none of which had had significant caries, had been extracted at multiple episodes of abscesses. At the time of examination he was in good health with normal stature and no physical abnormalities. He was the middle child with two dentally normal and healthy siblings. No other member of his family had any identified dental anomalies.

Gestation had been normal with full-term delivery, however at birth a flat and reddish birthmark under the right side of the lower lip had been noted. The medical history revealed no contributory serious illnesses, other than antibiotic therapy for 2 weeks for an ear infection at 2 years of age.

### Examination

Extraoral examination revealed a nonprominent birthmark on the facial skin under the right lower lip. Intraoral clinical examination revealed a caries-free, morphologically normal, full complement of primary maxillary teeth. All of the mandibular primary teeth were missing except the left canine and right second molar (Fig 1). The primary mandibular left canine had rough, hypoplastic yellow enamel and was moderately mobile. The primary mandibular right second molar had normal morphology with mild enamel hypoplasia. Full-mouth periapical radiographs and a panoramic radiograph (Fig 2) were obtained. Dental development from radiographs appeared age-appropriate and showed normal thickness of enamel and dentin in maxillary primary and permanent dentitions. However, excessive pulp horns and pulp stones were observed in some primary maxillary teeth. The radiographs indi-

cated ghost teeth for all permanent mandibular teeth except the right first molar. These ghost teeth were small, with thin layers of enamel and dentin, short roots, and wide pulp chambers. The mandibular left second premolar appeared to be missing.

### Treatment

Treatment goals consisted of the establishment of normal chewing function and improved esthetics and space management. The primary mandibular right second molar and left primary canine received pulpotomies and stainless-steel crowns to prevent pulp infection from excessive abrasion.

Alginate impressions were obtained for partial denture construction. The missing teeth were fabricated in wax to establish optimal occlusion and esthetics on the study cast. The wax-up model was duplicated and used to manufacture a template. The custom-made teeth were fabricated by pouring tooth-colored acrylic resin into the template (complete instructions for the fabrication of custom-made artificial teeth are available on request from the author). After the partial denture was made, a layer of resin (approximately 0.5 mm) was removed on the tooth-bearing portion of the denture and relined with soft liner material. Because there were no undercuts on the remaining primary mandibular teeth, wire clasp retainers were not practical. A thin layer of interim resilient liner was added to surround the primary canine and the second molar for improved retention. Additionally at delivery, a thin layer of soft liner was added to the tissue side of the denture to further improve retention.

The patient and parents were satisfied with the esthetics, function, and retention of the denture at delivery and at each 3-month follow-up appointment (Figs 3a and 3b). At the 1-year follow-up, the mandibular

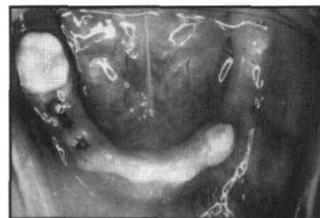


Fig 1. Intraoral photograph of the mandibular dentition.

right first permanent molar was found to have erupted with normal morphology. The denture was modified with a reinforced wire, self-cured acrylic resin to the right lingual posterior border of the denture, and a ball

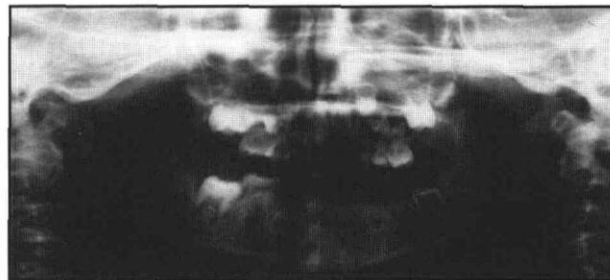


Fig 2. Panoramic radiograph showing normal maxillary dentition and affected mandibular teeth.

clasp between the right second primary molar and the first permanent molar. The modified denture exhibited greatly improved retention and stability.

## Discussion

The diagnosis of ROD is based on characteristic clinical and radiographic features. This patient presented with an intact primary maxillary dentition but premature loss of multiple primary mandibular teeth. The clinical findings were insufficient to support the diagnosis, as only two primary mandibular teeth remained with enamel hypoplasia. However, the roentgenographic examination (Fig 2) showed the distinct characteristics of ROD. The generalized ghost teeth features involving the mandibular permanent dentition (except the first permanent molar) strongly supported the diagnosis of ROD.

The etiology of ROD has been proposed by several theories. There was no contributory medical or trauma history for the patient described in this article. The patient was referred to an endocrinologist who reported no evidence suggestive of hypothyroidism or growth hormone deficiency and no other abnormal findings except ROD. Heredity did not appear to be a factor as no family members were identified with dental anomalies. Because the maxillary arch and mandibular right first permanent molar appeared unaffected, it is unlikely that this case is the result of a generalized structural disorder. Interestingly, several cases of ROD have been reported with a birthmark occurring on the affected

side suggesting a local vascular defect. This patient was born with a birthmark under the lower lip adjacent to the affected quadrants, supporting the theory of an underlying vascular etiology.

Most ROD cases present with dental abscess, jaw swelling, or gingival enlargement. Pinkham and Burkes<sup>8</sup> reported that the extraction of teeth with odontodysplasia is not the only treatment option. If the permanent teeth erupt, it is advisable to protect them from fracture and infection by restoring them with preformed crowns (without tooth preparation) to preserve the vitality of the affected teeth and permit continuing dentin formation. We also suggest preventive endodontic treatment (e.g., pulpotomy or partial pulpectomy) for the affected teeth to prevent pulp exposure from abrasion or mechanical preparation. In primary teeth, a pulpotomy with restoration using stainless-steel crowns should be performed as soon as the affected teeth erupt. For permanent dentition, pulpotomy or apexification<sup>9</sup> (depending on tooth vitality) should be considered to stimulate root formation. However, if the affected permanent teeth erupt with very short roots and wide-open apices, extraction may become necessary due to pulp necrosis and/or tooth instability.

The prognosis of the affected, malformed permanent teeth is poor due to the thin tooth structure, short roots, and open apices. Dental implants are a likely long-term treatment to construct a stable fixed or removable prosthesis. The timing of implant placement according to the growth and development of the dental arches must be considered.<sup>10</sup> Without adequate studies to support the use of implants in growing children,<sup>11-13</sup> they are not recommended for the case discussed in this article.

Though few of the case reports<sup>1</sup> have described treatment for ROD, it is generally treated by extracting the affected teeth and constructing a prosthetic replacement. Melamed, et al.,<sup>9</sup> however, presented a conservative multidisciplinary approach to treatment of a ROD case in the mixed dentition. This article proposed a treatment and restorative technique for ROD involving multiple primary teeth. Prosthetic restoration with partial dentures is important for these patients to provide chewing function, normal vertical dimension, space preservation, and improved esthetics. The use of a removable partial denture with soft liner aids retention by surrounding the supporting teeth, which lack undercuts for wire clasps. Custom-made artificial teeth provide better esthetics and occlusion than preformed denture teeth. Furthermore, due to the growth of alveolar bone and eruption of permanent teeth, 4- to 6-month follow-up appointments are important for periodic replacement of the soft liner and adjustment of the denture.

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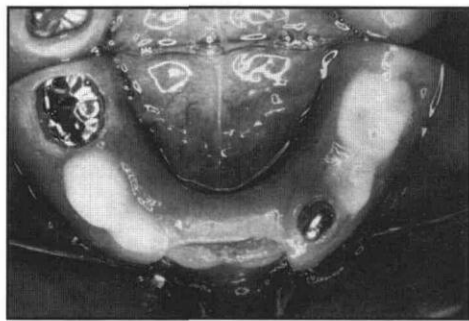


Fig 3a. Occlusal photograph of the mandibular partial denture in place.



Fig 3b. Frontal photograph of the partial denture in occlusion.

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