Clinical Management of Regional Odontodysplasia

Abel Cahuana, PhD, MD, DDS Yndira González, DDS Camila Palma, DDS

Dr. Cahuana is associate doctor, Pediatric Dentistry and Orthodontics Service, Hospital Sant Joan de Déu; Drs. González and Palma are graduates in dentistry and masters in pediatric dentistry, University of Barcelona, Barcelona, Spain.
Correspond with Dr. Cahuana at acahuana@hsjdbcn.org

Abstract

Regional odontodysplasia (ROD) is a rare, localized developmental anomaly of the dental tissues with distinctive clinical, radiographic, and histologic findings. The purpose of this study was to describe the characteristics and clinical management of 2 patients diagnosed with ROD at the Pediatric Dentistry Service at the Hospital Sant Joan de Déu, Barcelona, Spain. In both cases, temporary and permanent dentition were involved. It was concluded that therapeutic decisions during childhood must be based on the degree of involvement and each case's functional and aesthetic needs. Autotransplantation may be a good partial treatment option during the period of mixed dentition in some cases. Definitive treatment will include prosthetic rehabilitation with implants once the patient reaches adulthood. (*Pediatr Dent.* 2005;27:34-39)

KEYWORDS: ODONTODYSPLASIA, GHOST TEETH, TOOTH AUTOTRANSPLANTATION

Received July 15, 2003 Revision Accepted November 24, 2004

Regional odontodysplasia (ROD) is a relatively rare localized developmental anomaly of the dental tissues with specific clinical, radiographic, and histologic characteristics. Crawford[1] ascribed the first report of ROD to Hitchin in 1934, while others[2-6] suggested that McCall and Wald were the first to describe this condition in 1947. It was not until 1963, however, that Zegarelli et al coined the term "odontodysplasia" and Pindborg added the prefix "regional" in 1970.[1]

In an extensive review of ROD in 1989, Crawford and Aldred[1] stated females are more often affected than males (1.4:1) and that there was no association with race. These authors listed several etiological factors such as local circulatory disorders, viral infections, pharmacotherapy during pregnancy, facial asymmetry, local trauma, metabolic disturbances, somatic and neural mutations, and syndromal involvement. They also suggested that a combination of factors might be involved. In spite of this, ROD's etiology remains undetermined.

The criteria for ROD diagnosis are based on clinical, radiographic, and histologic findings. The maxilla is typically affected twice as often as the mandible.[7] Clinically, the condition is usually unilateral and rarely crosses the midline; exceptions are, however, occasionally found.[6-8] In most cases in which the damage crosses the midline, only the central incisor on the opposite side is affected.[7,9,10] The teeth are likely to be small, brown, grooved, and hypoplastic.[1,11] Eruption failure or delay is frequently seen[1,9,12-15] as well as abscesses or fistulae in the absence of caries.[1,7,16]

Radiographically, there is a lack of contrast between the enamel and dentin, both of which are less radiopaque than unaffected counterparts.[1] Moreover, enamel and dentin layers are thin, giving the teeth a "ghost like" appearance.[17] The pulp chambers are large, and usually present areas of relatively radiodense tissue (pulp stones or denticles).[18] The follicles of unerupted teeth are enlarged.[5]

ROD's histopathologic characteristics are distinctive. Typically observed are areas of cellular dentin, together with amorphous areas, generally concentrated within the coronal dentin of the affected teeth—which appear grey in sections stained with hematoxylin and eosin.[19] Extensive regions of interglobular dentin are also frequently described.[5,18] The enamel shows hypoplastic and hypocalcified areas, though the enamel closer to the dentinoenamel junction appears more normal.[20] Invaginations extending from the enamel surface to the dentin have been observed,[17] which may be the cause of the entry of bacteria, leading to pulpitis, pulpal necrosis and abscess formation.[1,16]

Despite severe tooth alterations, the bone itself is not affected[21] and retention of teeth with ROD does not interfere with normal alveolar development.

In a review of the literature through March 2003, 56 articles on ROD have been published, with a total of 138

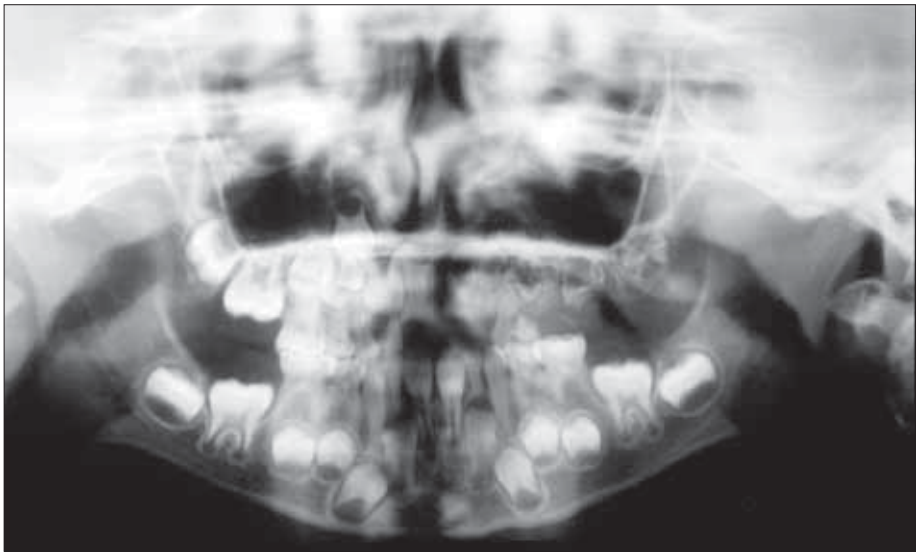


Figure 1. Panoramic radiograph of the patient at age 5 showing involvement of the upper right quadrant.

cases (including 31 since Crawford's last revision in 1989). The purpose of this paper was to examine some therapeutic considerations of ROD on the basis of 2 clinical cases.

Description of cases

Case 1

A 5-year-old boy, came to the Pediatric Dentistry Service at the Hospital Sant Joan de Déu of Barcelona due to pain and inflammation of the right maxilla. The parents reported a previous history of infections in this zone. The patient's medical history was noncontributory. Both parents reported no previous history of tooth or genetic anomalies on either side of the family.

Oral examination confirmed gingival swelling of the maxilla's upper right side of the maxilla, which was painful upon palpation. Moreover, the primary maxillary right molars and canine revealed an abnormal morphology, irregular surface, yellowish-brown color, and absence of caries. The primary maxillary right central and lateral incisors were fragmented up to the root level due to trauma. The morphology of the teeth in the other arches was normal, with the exception of the primary maxillary left central incisor which was avulsed when the patient was 14 months old.

Radiographically, both the primary teeth and follicles of the permanent teeth in the upper right quadrant had a "ghostly appearance." The enamel and dentin radiodensity of the permanent tooth buds were very similar; for this reason it was almost impossible to distinguish one tissue from the other. The pulp chambers were large. These alterations were compatible with regional odontodysplasia (Figures 1 and 2). A subgingival scaling procedure was performed on that visit. One week later, the patient was asymptomatic and gingival appearance was near normal. Due to the parents' concerns, the patient was placed on a 3-month recall schedule.

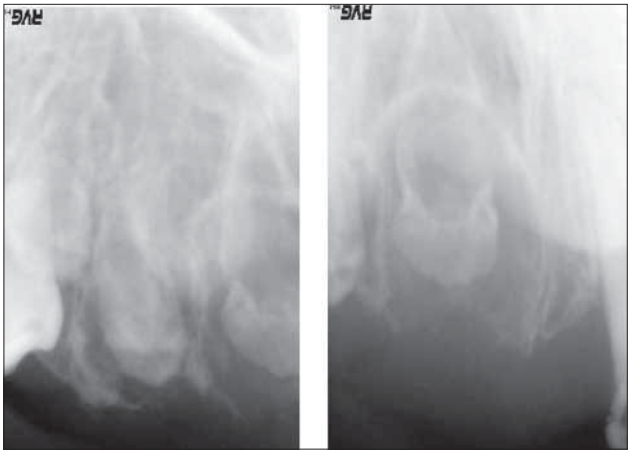
Two months later, however, the patient was treated for an emergency due to gross gingival enlargement and spontaneous pain. Consequently, the decision was made to extract the affected primary teeth and restore the missing teeth with an acrylic removable appliance covering from C to 3 (Figure 3). This appliance was administered to improve retention and prevent overeruption of the opposite teeth. This treatment allowed the dentition to remain stable. At a recall appointment, the first tooth of the adjacent quadrant (permanent maxillary left incisor) was found to be slightly affected.

Figure 2. Periapical radiograph showing regional odontodysplasia's characteristically "ghostly" appearance.

Figure 3. Acrylic removable appliance that was modified upon the eruption of teeth.

Figure 4. Clinical appearance of autotransplanted premolars.

Figures 5 and 6. Radiographic appearance of autotransplanted premolars with obliterated pulp chambers and healthy periodontal tissues.



Figures 7 and 8. Radiographic appearance of unerupted teeth nos. 6, 7 and 8.

Figure 9. Occlusal view showing teeth I and J with abnormal morphology and color.

At age 10, a dentoalveolar discrepancy in the 3 unaffected quadrants was observed, so the possibility of autotransplantations of premolars was considered. The premolars and permanent first molar of the maxillary right quadrant were surgically removed under general anesthesia. Immediately afterwards, the first premolars of the 3 other quadrants that had not yet completed their root formation (Nolla stage 7)[22] were autotransplanted into the right maxilla. The patient continued to use an acrylic removable appliance which was modified on the eruption of the autotransplanted teeth.

At age 15 during the last recall appointment, the premolars autotransplanted 5 years previously exhibited healthy soft tissues and occluded satisfactorily. There was a posterior crossbite, however, and the position of teeth was not ideal (Figure 4). The radiographs also showed normal periodontal ligaments and crestal bone level, although the pulps were obliterated (Figures 5 and 6). In the anterior area, the permanent maxillary right incisors and canine had not erupted (Figures 7 and 8). In the posterior area, the developing maxillary third permanent molar appeared to be less affected. Currently, the aim is to maintain the teeth of the anterior area until craniofacial development is completed and then perform prosthetic rehabilitation.

Case 2

A 3-year-old girl, presented at the Pediatric Dentistry Service at the Hospital Sant Joan de Déu, for an abscess in the upper left maxilla. Her medical history was noncontributory. The parents reported no previous history of the family's condition. Clinically, the primary maxillary left first and second molars were smaller than the other molars and presented an irregular surface, a yellowish-brown color, and absence of caries (Figure 9). The gingiva was swollen and tender on palpation. Radiographically, the primary maxillary left molars and maxillary left premolars and permanent molars had the "ghostly" appearance characteristic of ROD (Figure 10). Proper oral hygiene and antibiotic therapy were prescribed for 7 days to alleviate the symptoms and control infection.

At the recall visit 1 week later, there was no evidence of the infection and a total recovery of the gingival tissues was

observed. It was decided to follow a conservative treatment with oral hygiene instructions with frequent controls and maintain the affected primary teeth in situ.

At age 7, the patient presented the primary maxillary left second molar's roots and a failure of eruption of the permanent maxillary left first molar. Radiographic examination confirmed the involvement of the permanent maxillary left first molar which had not completed its root formation (Figure 12). An acrylic removable appliance was inserted (Figure 13).

Figure 10. Panoramic radiograph showing affection of the upper left quadrant.

The patient will continue to be seen regularly, and treatment decisions will be made in the future once the involvement of the neighboring teeth is confirmed.

Figure 11. Clinical view showing absence of infection in the upper left quadrant in spite of the coronal destruction of I and J.

Discussion

ROD is a relatively rare, nonhereditary, localized developmental anomaly of the dental hard tissues of a group of contiguous teeth. It occurs in both deciduous and permanent dentitions, but has a marked preference for the maxilla.[7] The etiology remains unknown, although several causative factors have been proposed.[1] Both cases described here cannot be related to any of the etiological factors previously described. Hence, an idiopathic origin is suggested. Diagnosis was based on the clinical and radiographic findings characteristic of ROD.

In both cases, only one quadrant in the maxilla was affected. In case 1, however, a slight degree of ROD was found in the tooth next to the affected quadrant, as has been previously reported.[9,10]

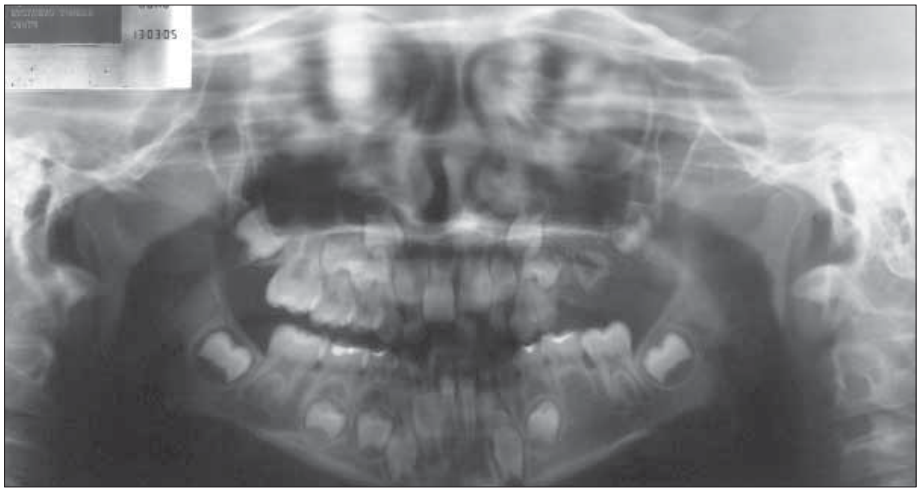


Figure 12. Affection of the maxillary left permanent first molar is confirmed radiographically.

ROD treatment remains somewhat controversial.[3,10,14] These cases require a continuous and multidisciplinary approach.[16,23] In a child with ROD, conservative treatment should be applied to preserve the affected teeth for as long as possible to provide normal jaw development.[3,7,24] Several reports[7,21,24] state that if abscessed teeth are present, they should be extracted and edentulous areas should be restored with acrylic removable appliances to:

1. maintain aesthetic and masticatory functions;
2. avoid overeruption of opposing teeth;
3. achieve space preservation and normal vertical dimension;
4. lessen the psychological effects of premature tooth loss.

clinical section

Figure 13. Acrylic removable appliance which avoided overeruption of opposite molars.

The conservative approach applied in case 2 (oral hygiene instructions and antibiotic therapy for a week), however, allowed the preservation of the affected primary teeth for 4 years without infection recurrence and pain.

Other reports have emphasized the use of the removable partial denture as part of the ROD treatment.[2,8,11,13,25]

As the bone itself is not affected by ROD, autotransplantation offers a good alternative if suitable donor teeth are available.[21] Autotransplantation is an accepted therapeutic option in dentistry[26,27] and has been successfully used to treat ROD.[21] Despite the increasing use of osseointegrated implants in patients with missing teeth, their use is contraindicated in growing patients.[28] Implants are preferably placed after pubertal growth.[10] The autotransplantation was considered in case 1 due to the dentoalveolar discrepancy in the 3 unaffected quadrants. The posterior site was preferred for autotransplantation because, at that moment, the permanent incisor had a better prognosis for eruption.

The posterior site allowed the extracted premolars to be placed in a natural alveolar bed corresponding to the affected teeth immediately after their extraction, without removing a considerable amount of bone. Moreover, this procedure preserved the posterior bone height and avoided the loss of vertical dimension.

A risk of ankylosis in autotransplanted teeth has been previously reported[26,27] and is diagnosed during the first year by radiographic appearance (loss of lamina dura) and a "high metallic percussive sound."[28] During the 5-year follow-up period of case 1, there was no evidence of ankylosis in the autotransplanted premolars (Figures 5 and 6). Nevertheless, prosthetic treatment with implants will be considered once the patient's craniofacial development is completed.

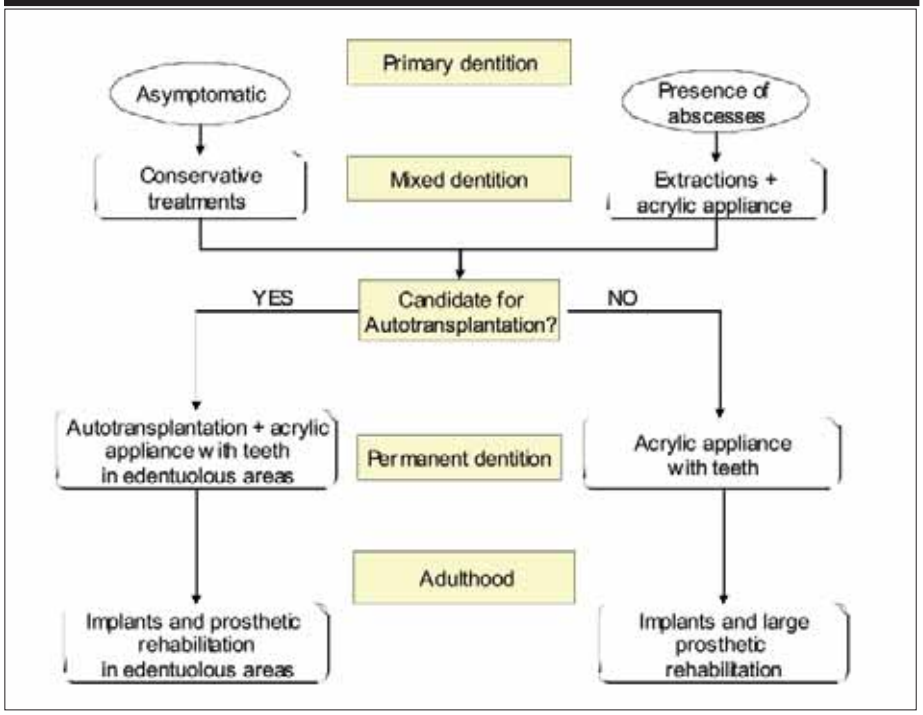
Conclusions

The treatment plan for ROD should be based on the degree of ROD, characteristics of unaffected areas, and the aesthetic and functional needs of each case. Individual management is required until the patient reaches the age for prosthetic rehabilitation. (The therapeutic considerations in cases of ROD are illustrated in Table 1.)

References

1. Crawford PJM, Aldred MJ. Regional odontodysplasia: A bibliography. J Oral Pathol Med 1989;18:251-263.
2. Oncag O, Eronat C, Hakan B. Regional odontodysplasia: A case report. J Clin Pediatr Dent 1996;21:41-46.
3. Kahn MA, Hinson RL. Regional odontodysplasia: Case report with etiologic and treatment considerations. Oral Surg Oral Med Oral Pathol 1991;72:462-467.
4. Neupert EA III, Wright JM. Regional odontodysplasia presenting as a soft tissue swelling. Oral Surg Oral Med Oral Pathol 1989;67:193-196.
5. Kinirons MJ, O´Brien FV, Gregg TA. Regional odontodysplasia: An evaluation of three cases based on clinical, microradiographic, and histopathological findings. Br Dent J 1988;165:136-139.

Table 1. Therapeutic Considerations in ROD



6. Guzman R, Elliot MA, Rossie KM. Odontodysplasia in a pediatric patient: Literature review and case report. Pediatr Dent 1990;12:45-48.
7. Lustmann J, Klein H, Ulmansky M. Odontodysplasia: Report of two cases and review of the literature. J Oral Surg 1975;39:781-793.
8. Sabah E, Eden E, Unat T. Odontodysplasia: Report of a case. J Clin Ped Dent 1992;16:115-118.
9. Williams AS, High AS: Odontodysplasia associated with orbital coloboma. Br Dent J 1988;164:390-394.
10. van der Wal JE, Rittersma J, Baart JA, van der Waal I. Regional odontodysplasia: Report of three cases. Int J Oral Maxillofac Surg 1993;22:356-358.
11. Yuan SH, Liu PR, Childers NK. An alternative restorative method for regional odontodysplasia: Case report. Pediatr Dent 1997;19:421-424.
12. Schmid-Meier E. Unilateral odontodysplasia with ipsilateral hypoplasia of the mid-face: A case report. J Maxillofac Surg 1982;10:119-122.
13. Pruhs RJ, Simonsen CR, Sharma PS, Fodor B. Odontodysplasia. J Am Dent Assoc 1975;91:1057-1066.
14. Sadeghi EM, Ashrafi M. Regional odontodysplasia: Clinical, pathologic, and therapeutic considerations. J Am Dent Assoc 1981;102:336-339.
15. Hanks PA, Williams B. Odontodysplasia: Report of two cases. Pediatr Dent 1988;20:199-203.
16. Vaikuntam J, Tatum NB, McGuff HS. Regional odontodysplasia: Review of the literature and report of a case. J Clin Pediatr Dent 1996;21:35-40.
17. Rushton MA. Odontodysplasia: Ghost teeth. Br Dent J. 1965;119:109-113.
18. Herold RCB, Lally ET, Gold L. Abnormal tooth tissue in human odontodysplasia. J Oral Surg 1976;42:357-365.
19. Gardner DG, Sapp JP. Ultrastructural, electron-probe, and microhardness studies of the controversial amorphous areas in the dentin of regional odontodysplasia. J Oral Surg 1977;44:549-559.
20. Kerebel B, Kerebel LM. Enamel in odontodysplasia. J Oral Surg 1981;52:404-410.
21. von Arx T. Autotrasplantation for treatment of regional odontodysplasia: Case report with 6-year follow-up. Oral Surg Oral Med Oral Path Oral Radiol Endod 1998;85:304-307.
22. Nolla CM. The development of the permanent teeth. J Dent Child 1960;27:254-266.
23. Melamed Y, Hornick J, Becker A, Shapira J. Conservative multidisciplinary treatment approach in an unusual odontodysplasia. J Dent Child 1994;61:119-124.
24. Lopes Marques AC, Castro WH, Vieira do Carmo MA. Regional odontodysplasia: An unusual case with a conservative approach. Br Dent J 1999;186:522-524.
25. Gomes M, Modesto A, Cardoso A, Hespanhol W. Regional odontodysplasia: Report of a case involving two separate areas. J Dent Child 1999;66:203-207.
26. Andreasen JO, Paulsen HU, Yu Z, Schwartz O. A long-term study of 370 autotransplanted premolars. Part II. Tooth survival and pulp healing subsequent to transplantation. Eur J Orthod 1990;12:14-24.
27. Andreasen JO, Paulsen HU, Yu Z, Schwartz O. A long-term study of 370 autotransplanted premolars. Part III. Periodontal healing subsequent to transplantation. Eur J Orthod 1990;12:25-37.
28. Thomas S, Dent M, Turner SR, Sandy JR. Autotransplatation of teeth: Is there a role? Br J Orthod 1998;4:275-282.

Abstract of the Scientific Literature

Attention Deficit Hyperactivity Disorder as a Risk Factor for Dental Caries

The aim of this article was to investigate if children with attention deficit hyperactivity disorder (ADHD) have higher dental caries experience than children without this condition. The 2 investigated groups consisted of: (1) 128 children with a DMFT score≥5 (case group); and (2) 128 children with a DMF score≤4 (control group). Identification of ADHD children was done by reviewing their medical history contained in the dental records and sending a questionnaire to their parents. The investigators also inquired for fluoride ingestion history, oral hygiene, and diet. A total of 9% of children in the case group and 2% of children in the control group had ADHD. Children medicated for ADHD were more likely to be in the high-disease group than in the low-disease group. After adjusting for preventive dentistry history, medical problems, and diet, ADHD children had over 10 times the odds of being in the high-disease group.

Comments: ADHD children may be at higher risk for dental caries. Therefore, pediatric dentists should reinforce preventive measures with them. More studies should be conducted to find the factors that may be increasing caries risk for ADHD children. JLC

Address correspondence to Dr. K.M.S. Ayers, Department of Oral Sciences, Faculty of Dentistry, University of Otago, PO Box 647, Dunedin, New Zealand.

Broadbent JM, Ayers KMS, Thomson WM. Is attention deficit hyperactive disorder a risk factor for dental caries? Caries Res 2004;38:29-33.

20 references