

# Regional Odontodysplasia: A case report

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## ABSTRACT

Regional odontodysplasia is a rare developmental anomaly affecting all germ layers. It may affect the primary or permanent dentition of the maxilla and/or mandible. The present case report describes the clinical, radiographic and histologic features and management of a 12 year old girl diagnosed with regional odontodysplasia.

**Key words:** regional odontodysplasia, dentin dysplasia

## INTRODUCTION

Regional odontodysplasia is a rare non-hereditary developmental dental defect with specific clinical and radiographic characteristics. It has been described under a variety of terms such as ghost teeth, regional odontodysplasia, odontogenesis imperfecta, non hereditary segmental amelogenesis imperfecta, unilateral dental malformation and odontogenesis dysplasia.<sup>1</sup> Regional odontodysplasia most commonly affects the anterior teeth with a slight female predilection.<sup>2</sup> The maxillary arch is involved twice as often as the mandibular arch.<sup>3</sup>

It affects both the primary and permanent dentition. Affected primary dentition is usually succeeded by affected permanent dentition.<sup>3</sup> However, affected permanent teeth may succeed normal primary dentition and rarely normal permanent dentition follows affected primary ones.

The diagnostic criteria for regional odontodysplasia (Crawford and Aldred) are predominantly clinical and radiographic although the histopathologic findings are



Fig 1



Fig 2

also distinctive.<sup>3</sup> Enamel and dentin formation is defective with enamel showing irreg-

ular surface and dentin. The radiographic appearance is unique because of marked reduction in radiodensity. The etiology remains uncertain and treatment may sometimes be controversial.

The present case report describes a case of regional odontodysplasia in a 12 year old female child.

## CASE REPORT

A 12 year old healthy, girl hailing from Pakistan reported to Subharti Dental College, Meerut, Uttar Pradesh, India with chief complaint of non-eruption of anterior right maxillary permanent teeth. Her prenatal, birth, medical and family histories were unremarkable. Clinically, tooth #11, 12, 13 and 53 (Figure 1) were missing. Radiographic examination (Intraoral periapical radiograph) revealed presence of unerupted tooth #11, 12 and 13 (Figure 2) with retarded dental development for the



age of the child. The teeth showed decreased radioopacity with indistinct enamel dentin demarcation. The roots were short, blunt and underdeveloped. On the basis of clinical and radiographic examination a provisional diagnosis of regional odontodysplasia was made. Under local anaesthesia, all the unerupted right maxillary permanent teeth were surgically extracted and sutures placed (Figure 3).

The extracted teeth (Figure 4) showed altered morphology with yellowish-brown color and short/undeveloped roots. The specimen, consisting of teeth fragments, was sent to for histopathologic examination which gave a clinical diagnosis consistent with that of regional odontodysplasia.

Histologically, the ground section revealed normal appearing enamel. The underlying dentin was thin and irregular with broad areas of interglobular dentin. The predentin also contained osteodentin. The dentino-enamel junction was not well scalloped. The pulp chambers were large with the presence of pulp stones. A thin layer of acellular cementum with incremental lines was present.

Two weeks postoperatively, an acrylic maxillary removable partial denture was constructed and inserted to preserve the alveolar ridge during the period of skeletal growth. This was followed by oral hygiene maintenance instructions the patient was placed on recall to monitor the development of the maxillary arch.

## DISCUSSION

Regional odontodysplasia is a developmental anomaly affecting dental tissues of both ectodermal and connective tissue origin. It most commonly involves the maxillary arch (with a 2:1 maxillary-mandibular arch frequency ratio) and has no specific racial predilection.<sup>2,3</sup> Although the average age of diagnosis is not specific, regional odontodysplasia commonly manifests at the time of eruption of primary or mixed dentition.<sup>4</sup> The condition is commonly unilateral (as in our case) although occasional cases do cross the midline.<sup>2</sup> Cases in which the defect crosses the midline may be referred to as generalized odontodysplasia.<sup>4</sup> Rarely, almost all teeth of the same arch are affected. Cases have also been

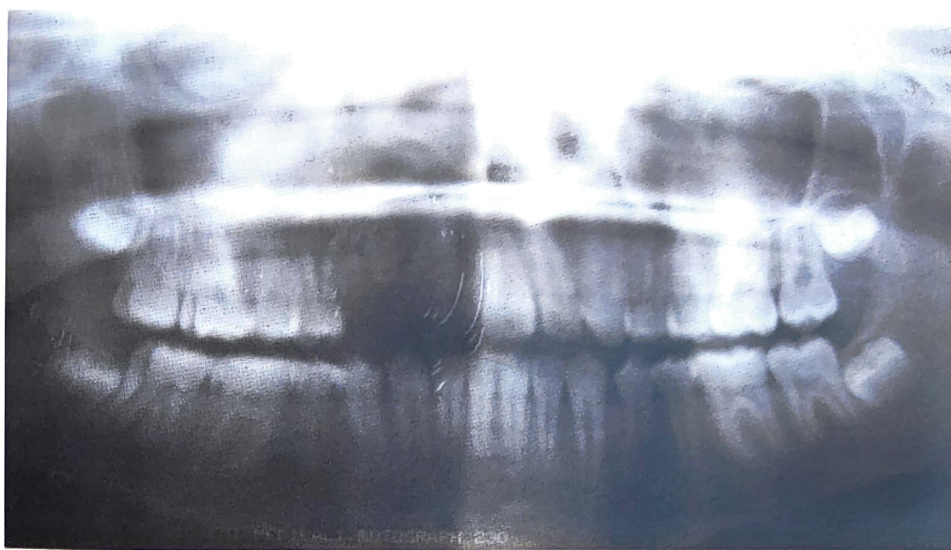


Fig 3

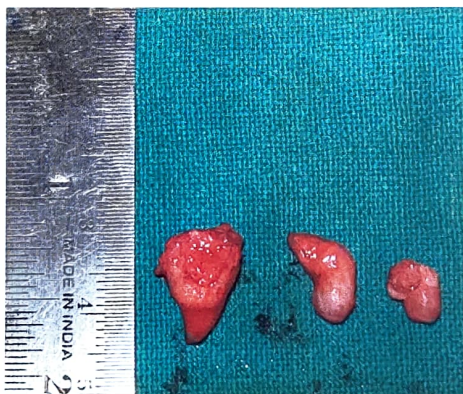


Fig 4

reported with involvement of both arches.<sup>5</sup> Teeth affected with regional odontodysplasia are generally seen as a continuous series.<sup>5</sup> However, in some cases the anomaly may 'skip' a tooth or group of teeth.<sup>6</sup> The central and lateral incisors and canines are most commonly affected in maxilla or mandible.<sup>5</sup> In our case, the right maxillary quadrant was affected unilaterally, in a 12 year old female child.

Histologically, all structures of the dental germ are affected.<sup>7</sup> Enamel formation is defective with enamel characterized by decreased thickness, irregular morphology and hypomineralization with focal areas of dentin exposure. Dentin is thin and hypomineralized wide predentin zone, fewer dentinal tubules and prominent interglobular dentin. Cellular dentin and

amorphous areas within coronal dentin are considered to be pathognomonic for regional odontodysplasia.<sup>3,7,8</sup> The pulp chamber is large and pulp necrosis is often present.<sup>5</sup> Teeth which develop late have a tendency towards more normal structure than those that develop early in life.<sup>8</sup> The histologic findings in our case were consistent with these features.

Radiographically, there is a lack of contrast between dental hard tissues in teeth affected by regional odontodysplasia as was seen in the present case. Both enamel and dentin of affected teeth seem less radioopaque than those of adjacent teeth. Enamel and dentin are thin, imparting a 'ghost-like' appearance to the tooth. The pulp chambers are large with calcifications and open apices at root end. In our case, the affected teeth # 11, 21 and 31 showed short roots with blunted apex and 'ghost-like' appearance.

The etiology of regional odontodysplasia is yet unclear and several possible causes have been reported in the literature including local trauma, somatic mutation, local ischemia, Rh incompatibility, activation of latent virus in odontogenic epithelium, local infection, neural disorders, failure of migration of neural crest cells, pharmacotherapy during pregnancy, irradiation and hyperpyrexia. Thus, whatever the specific pathogenesis, the causative factor must be active in both deciduous and permanent dentition. A local neural disorder



(failure of migration of neural crest cells) or local ischemia have been suggested to best account for the asymmetric localized nature of regional odontodysplasia. The chief complaint is usually delayed eruption or associated gingival inflammation, pain or abscess.<sup>2</sup> The patient in our case reported with the chief complaint of non-eruption of maxillary anterior permanent teeth, unaccompanied by gingival inflammation and/or abscess formation. The treatment of regional odontodysplasia remains controversial. The chief reason for extraction of the affected teeth since, even if they erupt, the teeth are defective and of undesirable appearance.<sup>4,8,9</sup> The longer the affected teeth are retained, the greater the chances of development of associated pathology.<sup>7</sup>

A variety of conditions have been reported to be associated with regional odontodysplasia. These include vascular abnormalities such as hemangiomas, unilateral hypoplasia of the zygomaticomaxillary complex, epidermal nevus syndrome and gingival enlargement.

Several factors must be considered to determine the suitable treatment options in a pediatric patient with regional odontodysplasia. These factors are<sup>7</sup>

1. Age of the patient
2. Presence of systemic pathology
3. Aesthetic and functional needs
4. Extent of regional odontodysplasia

Treatment for regional odontodysplasia is controversial. General aims of treatment include maintenance of aesthetics, reducing psychologic impact, phonation and masticatory function. Conservative treatment may be carried out for a child affected with regional odontodysplasia in order to preserve the affected teeth for as long as possible and encourage the normal growth and development of the jaws.<sup>4</sup> Autotransplantation has also been cited in the literature as acceptable treatment option for regional odontodysplasia.<sup>10</sup> However, if the affected teeth show associated pathology such as abscess formation or in light of the fact that these teeth, if erupted are usually of defective type, extraction should be considered.<sup>4,7</sup> This may be followed by restoration of the edentulous area created with a removable appliance as was done in our case.<sup>4</sup>

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**"Dental Motivation and Education Camp" for school teachers held under the initiative of Dr.I.K.Pandit in association with the ICD. The camp was held in a village based place at Siddhivanayak Group Of Colleges, some 15 kilometres away from DAV (C) Dental College Yamunanagar where a team of expert doctors explained the teachers of the school the importance of early reporting, early detection and prevention of dental diseases. Teachers training camp aimed at asking the teachers to include oral health education in their curriculum. The principal of the school was assured that the same awareness camps shall be a routine under the aegis of International College Of Dentists-India section.**

