Dört Yarım Çeneyi İçeren Atipik Rejyonel Odontodisplazi: Bir Olgu Raporu

Atypic Regional Odontodysplasia Involving Four Quadrants: A Case Report

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Özet

Rejyonel odontodisplazi nadir görülen, mine, dentin ve pulpanın herediter olmayan bir anomalisidir. Rejyonel odontodisplazinin etiyolojisi bilinmemektedir. Bu anomali genellikle ağzın sadece bir kuadranını içerir ve maksiller keser dişler sıklıkla etkilenen dişlerdir. Hipoplastik ve kahverenkli dişler rejyonel odontodisplazi için karakteristiktir. Eksik veya gömülü dişler bu patolojide sıklıkla görülmektedir. Radyolojik incelemede, geniş pulpa odalı, azalmış radyoopasiteye sahip "hayalet diş"görüntüsü mevcuttur. Rejyonel odontodisplazi ve amelogenezis imperfektanın benzer özellikleri nedeniyle klinik olarak ayrımlarını yapmak oldukça zordur. Ayırıcı tanı için klinik, histopatolojik ve radyografik değerlendirme birlikte yapılmalıdır. Diş yapısındaki hasar yüzünden ideal ve uzun ömürlü restoratif tedavi rejyonal odontodisplazi hastalarında sıkıntılı bir konudur.

Sunulan vaka raporunda, 21 yaşındaki kadın hastada dört kuadranı içeren atipik rejyonel odontodisplazinin tanı ve tedavisi anlatılmaktadır.

Anahtar Kelimeler: Rejyonel Odontodisplazi, dental anomali, hayalet diş, amelogenezis imperfekta

Abstract

Regional odontodysplasia is a rare anomaly of enamel, dentine and pulp tissue without any hereditary patern. The etiology of regional odontodysplasia is unknown. This anomaly generally involves only one quadrant of the mouth, and anterior maxillary teeth are mostly affected region. Hypoplastic and brown colored teeth are characteristic for regional odontodysplasia. Absent teeth or impacted teeth are also common with this pathology. Wide pulp chambers and less radiopaque teeth appearance is typical (ghost teeth) in radiographic examination. The clinical differentiation of regional odontodysplasia and amelogenesis imperfecta is really difficult because of their similar clinical features. Clinical, hystopathological and radiographic evaluation should be considered together for differential diagnosis. Because of the damaged tooth structure, ideal and long lasting treatment in patients with regional odontodysplasia is a distressing issue.

Present report describes diagnose and treatment of an atypical regional odontodysplasia case that involves four quadrant of the mouth in a 21 year-old female patient.

Key Words: Regional Odontodysplasia, dental anomaly, ghost teeth, amelogenesis imperfecta

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Regional odontodysplasia (RO) is a rare, non-hereditary developmental anomaly affecting soft and hard dental tissues derived from both mesoderm and ectoderm with typical clinical and radiographic appearance[1-4]. Many different terms such as odontodysplasia, ghost teeth, odontogenesis imperfecta, localized arrested tooth development, unilateral dental malformation, and familial amelodentinal dysplasia have been introduced to describe this pathology[1,4]. Odontodysplasia was first described by Zergalli et al. in 1963 and the term regional was added by Pindborg in 1970[5,6]. RO can occur both in primary and permanent dentition and generally involves only one quadrant. Involvement of more than two quadrants is very rare. It is more common in females and there is a maxillary predominance[1,4,7]. Circulatory disorders, trauma, latent virus in tooth germs, metabolic disturbances, local infections, somatic mutation, neural alterations, hypophosphatasia, hypocalcemia and irradiation are possible causes of this pathology[1,4,7]. Although RO is described as an isolated anomally; it can be accompanied with developmental anomalies such as unilateral facial hypoplasia, neurofibromatosis, vascular nevi, mental impairment, ectodermal dysplasia, Papillon-Lefevre and Gorlin syndromes[3]. In some cases facial asymmetry may also be seen with RO[8]. The most common complaints of these patients are absent teeth or delayed tooth eruption, dental abscesses and poor dental esthetics[1,2]. Affected teeth are small, yellowish-brown colored and have irregular surface with pitting and grooves[4,7]. Enamel layer is also soft on probing[9]. When RO affects the primary dentition, teeth eruption does not delay but, gingiva becomes hyperemic and generally fistula formation occurs. However in permanent dentition; affected teeth are usually impacted or partially erupted and gingival swelling occurs[4]. In radiography, enamel and dentin layers are thin with reduced radioopacity and the demarcation line between these layers is not visible[4,7,9]. Histologically, hypocalcified enamel areas and impaired prism structure are visible[4,9]. Thickness of dentin layer is less than normal and interglobuler dentin areas are irregular formed and spread. Generally, pulp tissue contains dentickels and amorphous calcified material[7,9]. Increased pulp wideness is a characteristic feature of RO and it provides the ghost teeth appearance of the teeth. In this report an atypical and rare case of regional odontodysplasia which involves .

Case Report

A 21 year-old girl, who was complaining about absence of permanent teeth and esthetic distortion of yellowish-brown colored teeth referred to the clinic. She had no systemic disorder, trauma history and no medical or dental problem in family members.

A mild maxillary retrusion and reduced vertical height of lower third of the face were detected in extraoral examination of the patient. Intraoral examination revealed yellowish-brown colored, irregular surfaced, soft enamel layered microdontic teeth (figure 1). Numerous absent teeth were detected in all four quadrants. Some primary teeth (55, 63, 65 and 85) were still persistent with multiple dental caries and, hyperemic and edematous surrounding soft tissue. All erupted teeth were shown in table 1. (Table 1)

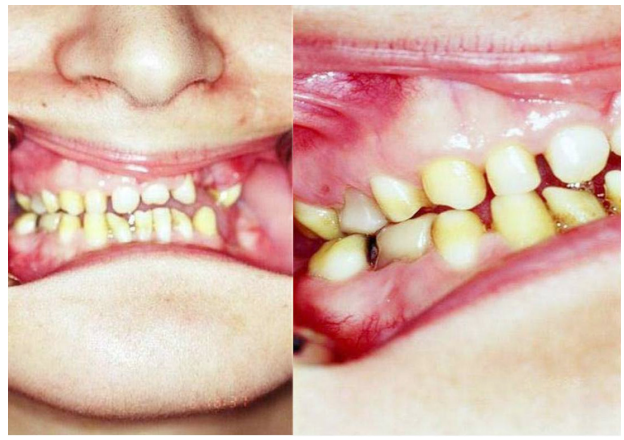

Figure 1a. **Figure 1b**.

Fig. 1a, 1b Preoperative introoral images showed microdontic, yellowish-brown teeth and edentulous regions

Table I. Shows the intra oral dental condition of the patient.

6 V 4 3 2 1	1 2 III V 6
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In panoramic radiography, numerous impacted teeth were detected in four quadrants. All impacted teeth were abnormally formed with short roots, wide pulp chambers, thin hypocalcified dentin and enamel layers and, showed characteristic "ghost teeth" appearance with reduced radioopacity (figure 2a, 2b).

Removal of all impacted and persistent primary teeth was planned and performed with local anesthesia in five sessions. Extracted teeth were evaluated histopatologically. In histological examination dysplastic cement, dentin-like calcifications and fibrosis in the pulp (figure 3a), irregular globular areas and abnormalities in channel structure in dentin (figure 3b) and dysplastic cement-like, odontogenic, mineralized hard tissues in-cement (periodontal tissue) (figure 3c) were observed.

Final diagnose was atypical regional odontodysplasia involving four quadrants based on clinical, radiographic and histopathologic findings. Following the extraction of all planned teeth, the rest of teeth were prepared and the temporary full mouth restoration was made. The temporary prosthesis was designed for functional rehabilitation and 3mm increasing of occlusal height. 6 months later, two implants were inserted to left posterior mandibular area (figure 4). 2 more months were waited for implant's osteointegration.

Finally, jaw relation registration was performed and the models were mounted on an articulator with present dimension of the vertical height of the occlusion. The final restorations were prepared based on these relation (figure 5).

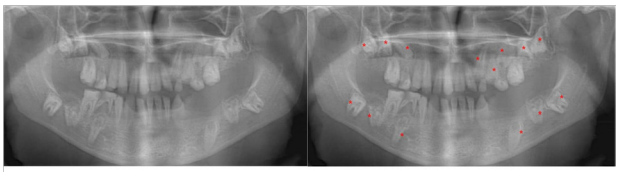

Figure 2a. **Figure 2b**.
Fig. 2a Preoperative panoramic radiograph of the patient illustrating teeth with RO.

Fig. 2b "*" signs were used to show teeth with typical "ghost teeth" appearance which is characteristic for RO.

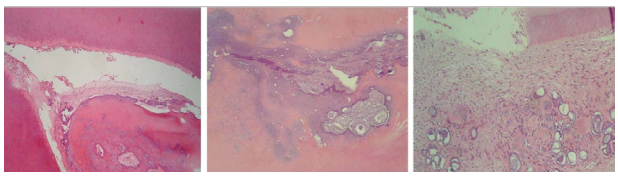

Figure 3a. **Figure 3b**. **Figure 3c**.

Fig. 3a Dysplastic cement, dentin-like calcifications and fibrosis were seen in the pulp. (HEX100)

Fig. 3b Irregular globular areas and abnormalities in channel structure were detected in dentin. (HEX100)

Fig. 3c Dysplastic cement-like, odontogenic, mineralized hard tissues were defined in periodontal tissue. (HEX100)

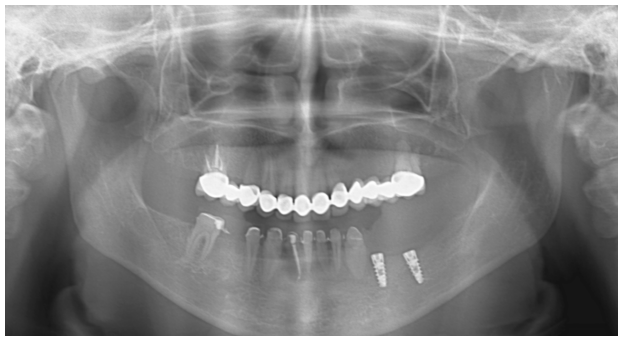

Figure 4.

Fig. 4 Panoramic view of the patient following the implant placement.

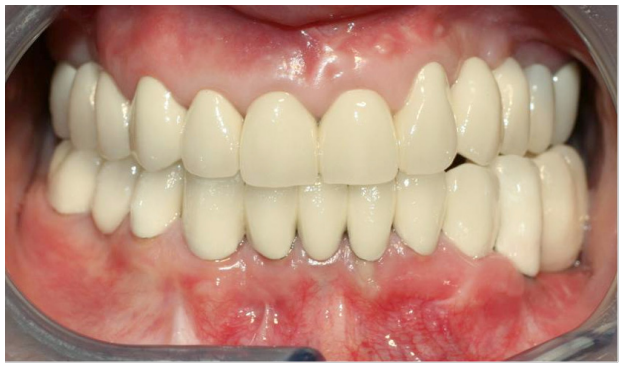

Figure 5.

Fig. 5 Final view of the patient with prosthetic restoration.

Discussion

RO is a rare developmental anomaly that affects every term of the teeth development and causes characteristic clinical, radiographic and histological findings[10, 11].

Although RO etiology is uncertain, several possibilities such as local circulatory disorders, latent virus on teeth germs, trauma, metabolic disturbances, medications during pregnancy, irradiation, genetic transmission have been suggested[3, 4, 12].

Most accepted etiology is local ischemia due to the vascular disturbances affecting tooth development[3, 10]. In the current case etiologic factor could not be identified. RO usually affects only one quadrant and the most common complaint is tooth impaction, delayed eruption or agenesis[4, 7]. However in the presented case, characteristic features of RO were seen in all four quadrants.

The differential diagnoses of RO are amelogenesis imperfecta (especially hypocalcified type), dentinogenesis imperfecta, dentinal dysplasia and hypophosphatasia [4, 13]. In this case the most controversial differential diagnosis was amelogenesis imperfecta because of its clinical features. Amelogenesis imperfecta is a hereditary (autosomal dominant or recessive to xlinked dominant or recessive) disorder of enamel formation that affects both dentitions[12]. The affected teeth are yellowish-brown colored and enamel layer is thin in amelogenesis imperfecta same as RO[14]. Although the clinical features of the presented case are similar with amelogenesis imperfecta, there was no familiar history of a dental anomaly in this patient. In contrast to RO, small pulp chambers are typical in amelogenesis imperfect[15].

In presented case pulp chamber extension was detected in all affected teeth. Irregular enamel layer resorption of unerupted teeth can also be seen in amelogenesis

imperfecta however all enamel, dentin and cement tissues of unerupted teeth are affected in RO[16]. Delayed eruption or impaction of permanent teeth was both reported with amelogenesis imperfecta and RO[17, 18]. However ghost teeth formation is characteristic for RO.

Removal of all impacted teeth was quite difficult because of their morphologic structure. They were extremely soft and broken into small pieces during the

elevation. RO was decided as precise diagnosis based on all this clinical, radiographic and histological findings. There is no appropriate treatment procedure for RO. Some clinicians prefer prosthetic rehabilitation following the removal of all affected teeth while others prefer to keep affected teeth as long as possible. If infection is absent, odontodysplasic teeth may be treated with restorations[4, 7]. Removal of odontodysplasic teeth affects alveolar bone formation and this may have a negative influence on the growth of facial skeleton[2]. In the presented case, giving a decision of treatment protocol was difficult because of wide ranges of tooth were affected. We preferred to keep all erupted permanent teeth, inserted two dental implants into acceptable bone areas after six months healing period and planned fixed prosthetic rehabilitation to provide the esthetic and functionional results. Multidisciplinary evaluation is necessary for providing the sufficient esthetic results and ideal function in patients with dental anomaly. The main complaint of this patient was poor esthetics. The extractions of persistent primary teeth were decided due to their insufficient functional support for long term prosthetic rehabilitation. All impacted teeth were surgically removed because of their structure and position. Erupted permanent teeth were kept and restorated for fixed prosthetic rehabilitation. Patient has effectively used her final prosthesis for twelve month.

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