

Regional odontodysplasia: a review of the literature and report of a case

MAHMOUD A. HAMDAN¹, FALEH A. SAWAIR², LAMIS D. RAJAB¹, AHMAD M. HAMDAN¹ & IYAD KH. AL-OMARI¹

¹Department of Paediatric Dentistry and Orthodontics, and ²Department of Oral Surgery, Oral Medicine, Oral Pathology and Periodontics, Jordan University Hospital, Amman, Jordan

Summary. We present a rare case of a developmental anomaly called regional odontodysplasia or ‘ghost teeth’ in an 8.5-year-old Jordanian girl. The anomaly affected both the mandibular primary and permanent incisors, and the canines bilaterally. The maxillary teeth were unaffected. This is the first case of the anomaly to be reported in Jordan. The clinical, radiographic and histological features are reviewed. The management of affected patients is discussed.

Introduction

Regional odontodysplasia (RO) is an uncommon, nonhereditary developmental anomaly involving components of both the dental mesoderm and ectoderm [1,2]. The condition can be differentiated from other odontogenic disturbances since all the histological elements of the dental organ are abnormal in the affected teeth, while other teeth in the same individual are normal [3]. Regional odontodysplasia has been reported under many other names, such as ‘ghost teeth’, ‘odontogenesis imperfecta’, ‘odontogenic dysplasia’, ‘nonhereditary segmental amelogenesis imperfecta’ and ‘unilateral dental malformation’ [4,5].

Review of the literature

Although Hitchin was the first to recognize this condition in 1934 [6], McCall and Wald [7] were credited for publishing the first report of odontodysplasia in 1947, in which they termed the condition ‘arrested tooth development’. In 1954, the term ‘shell teeth’ was introduced by Rushton [8], which the author used to describe the radiographic findings of the anomaly. In 1963, Zegarelli *et al.* [9]

were the first to suggest the term ‘odontodysplasia’. The term ‘regional’ was added because the condition affects a group of several adjacent teeth in a particular segment of the jaw.

The age of the patient at presentation is variable, although the condition typically manifests during the time of primary tooth eruption or during the mixed dentition [10]. Regional odontodysplasia is slightly more common in females and there is no tendency for its occurrence in any specific ethnic group [11]. Generally, the disturbance is localized to one arch and the maxilla is involved twice as often as the mandible [12]. The left side of the maxilla is the most frequently affected site followed, in order of decreasing frequency, by the maxillary right, mandibular right and mandibular left regions [12]. The number of affected teeth is variable and the affected teeth are usually in a continuous series. In the maxilla or mandible, the central and lateral incisors, and canines are the teeth most commonly affected [13]. In rare cases of RO, a single tooth has been reported to be affected [14], and the condition has sometimes been seen to ‘skip’ a tooth or a group of teeth [15]. The condition is usually unilateral, without tendency to cross the mid-line [11,16]. Not all cases of RO are ‘regional’: some authors have reported cases affecting both the maxilla and the mandible on the same side [13,17,18], and others have reported cases in which both sides of the same

Correspondence: M. A. M. Hamdan, University of Jordan, Faculty of Dentistry, Amman 11942, Jordan. E-mail: anwarm@ju.edu.jo

jaw were affected [3,19]. In extremely rare reports, RO affected all four quadrants [20]. These cases have been referred to by some authors as 'generalized odontodysplasia' [10]. In some cases, RO has been seen to cross the mid-line, particularly in the lower jaw [3,11,21]. If the primary teeth are affected, the permanent successors are usually affected as well [12]. However, affected permanent teeth may succeed normal primary teeth [22], and very rarely, normal permanent teeth follow affected primary ones [23].

Clinically, the affected teeth are atypically shaped with surface pits and grooves, hypoplastic, hypocalcified, and show yellowish or brownish discoloration [1]. Some of the affected teeth are whitish in colour at eruption, and later become yellowish or brownish [24]. Because their structure is defective, they are usually small in size and more susceptible to dental caries [4]. The alveolar crest in the region of the affected teeth is usually enlarged and covered by fibrous tissue [1]. The eruption of the affected teeth is behind schedule or does not occur at all [25,26]. When curetted, the unerupted teeth are extremely friable, and the dentine is very soft and could be mistaken for advanced caries [25].

Radiographically, the affected teeth show abnormal morphology and hypoplastic crowns. The enamel and dentine are less radio-opaque than unaffected counterparts, and there is little demarcation between enamel and dentine. This faint outline of the affected teeth was the reason for the term 'ghost teeth' [20,27]. The pulp chambers and root canals are wide, and the roots are short with wide and open apices [28]. Calcification is occasionally seen within the pulp chambers or root canals [17]. In some cases, the unerupted teeth are surrounded by a pericoronal radiolucency representing an enlarged dental follicle [29].

Histologically, all structures of the dental germ are affected. In ground section, the enamel is of variable thickness, producing an irregular surface [12]. The enamel prisms are irregular and the enamel may occasionally lack a prismatic structure [12], and it is hypoplastic and contains degenerated globular calcifications [30]. There is also hypocalcification of the enamel because residual enamel matrix is frequently seen in demineralized sections [31]. The dentine is thin, and the tubules are reduced in number and tortuous in shape [4]. Interglobular dentine and globular masses interrupting the dentinal tubules are frequently seen. Clefts within dentine,

some of which establish communication between the pulp and the oral cavity, are common findings [12]. Cellular dentine and amorphous areas within the coronal dentine are usually evident [15]. Closer to the dentino-enamel junction, the dentine is more evenly calcified [4]. Although the predentine is of variable thickness, it is usually wider than that seen in normal teeth [5]. Prominent interglobular dentine is also seen in the radicular dentine; however, the radicular dentine and cementum are generally less abnormal compared with the coronal dentine [32]. The pulp chamber is large with occasional long pulp horns, and often contains large irregular, calcified globules or stones [4]. Pulp necrosis is often noted as a result of the communication with the oral cavity through the dentinal clefts and pulp horns [15]. There is a strong trend for teeth which develop relatively late to have more a normal structure than those which develop early in life [3].

Dental follicles are composed of dense fibrous connective tissue [29,30,33]. Odontogenic epithelial rests, whorled fibrous tissue and foci of calcifications, occasionally coalescing into larger globose areas with an accentuated affinity to haematoxylin, are frequently seen within the dental follicle [12]. The enlarged gingiva which accompanies the affected teeth in some cases usually shows a parakeratinized surface epithelium with acanthosis and very hyperplastic rete ridges. The lamina propria is composed of fibrous connective tissue and contains chronic inflammatory cell infiltrate [4]. Calcified globules and odontogenic epithelial rosettes similar to those found in the dental follicle are also sometimes observed inside this tissue [4]. The calcifications are thought to be the result of: degenerative change of the reduced enamel organ [31]; degenerative change of the connective tissue cells [34]; earlier inflammation [35]; or formation by metaplastic epithelial cells [24].

In spite of its specific clinical, radiographic and histological features, RO is a condition of unknown aetiology [36]. The condition is not hereditary and it appears to be the result of local factors affecting the tooth-forming tissues during development [24]. Several factors have been suggested as causes in the literature, such as: the activation of a latent viral infection of the tooth germ during development; local trauma or ischaemia; irradiation; metabolic and nutritional disturbances; rhesus incompatibility; local somatic mutation; genetic transmission; medications taken during pregnancy; failure of

migration of the neural crest cells; and local vascular defects and haemangiomas [20,28,29,37,38]. However, no one factor has been positively identified as the single cause of the condition. In addition, it remains hard to explain why, in the vast majority of cases, particular contiguous teeth are affected with no involvement of others. There are occasional reports of relative normalization and development of the affected teeth in RO after a considerable delay [12,24,39], suggesting the cessation of the causative factor in these cases.

Although the histological features are distinctive, diagnosis is based on clinical and radiographic findings, and coincides with the eruptive periods of the primary and permanent teeth. Many of the cases are discovered incidentally during routine clinical and radiographic examinations [11,36]. The chief complaint is often delayed eruption or a non-inflammatory gingival swelling in the vicinity of the affected teeth [11]. Some patients present with pain and abscesses in the affected area [24] even in the absence of gross caries. These symptoms have been attributed to bacterial access to the pulp through clefts in the defective enamel and dentine [29]. The patient and/or parent may also present complaining of unpleasant appearance, problems in speech or mastication, draining fistulas, or facial asymmetry related to the loss of vertical dimension [4]. In some reports, affected patients also had vascular naevi [19], hypoplasia of the affected side of the face and facial asymmetry [37,40], hydrocephalus [41], dolichocephaly and clinodactyly [2], epidermal nevus syndrome [42], hypophosphatasia [43], and other abnormalities [12].

Although conditions such as dentinal dysplasia, amelogenesis and dentinogenesis imperfecta show some similarities to RO, these conditions affect the entire dentition without segmental involvement. When the affected teeth erupt, RO can be misdiagnosed as dental caries, and therefore, the teeth will be extracted without being submitted for histological examination [12]. Many cases of RO are also misdiagnosed as malformed teeth or odontomes [44,45].

Most dentists elect to extract the teeth involved immediately and later rehabilitate the patient with a temporary removable partial acrylic prosthesis because, even if they erupt, the teeth are defective and of undesirable appearance [3,10,13,44,45]. The longer the affected teeth are retained, the higher the chances of pathology developing. This will necessitate extraction of the teeth, and their removal may

be more difficult, especially if they are unerupted [5]. The temporary prosthesis can be maintained till the age of 17 or 18 years, a time when the gingival margin is stable and restoration with a fixed prosthesis can be considered.

Others clinicians have argued that removal of teeth at a young age may lead to undesirable psychological effects, and a substantial reduction in alveolar ridge height. The subsequent defect would also pose immense problems for future restorations should the patient want a fixed prosthesis or implants [1,36]. In addition, loss of the vertical dimension on the affected side might lead to defective jaw development and subsequent facial asymmetry [24].

Placement of osseointegrated implants in growing children with hypodontia is well documented in the literature [46–48]. Since the general quality of bone is not affected in RO, there may be a role for implants in such cases. Nevertheless, care must be taken when carrying out such procedures since there have been reports of a lower density of bone around affected teeth [16]. Care must also be taken to place mandibular implants forward of the mental foramen [48], and a certain amount of implant impaction is expected in the maxilla because of relocation of the maxillary sinus and nasal floor with growth [48].

One case of RO was followed radiographically for a 6-year period, during which time the ghost teeth exhibited significant dentin formation along with a resultant decrease in pulp size and relative normalization of the radicular anatomy [24]. Restorative procedures to protect the affected erupted teeth have been suggested [10,38,49]. Some clinicians have even suggested moving those teeth which have the most-developed roots orthodontically with subsequent restorations or prosthesis fixed on the pillar elements remaining [13].

Extreme caution must be exercised if movement of affected teeth is to be attempted. Since the roots of these teeth are short and a lower density of bone has been observed around affected teeth [16], forces used for orthodontic tooth movement must be kept very low to avoid both root resorption and undermining bone resorption. Furthermore, the atypical shape of the crowns of these teeth and their hypoplastic nature make it very difficult to bond orthodontic brackets to these teeth.

Once the permanent dentition has been established, joint orthodontic/restorative treatment aimed

at uprighting any unaffected teeth that are severely tilted into extraction spaces may be commenced. Treatment for other malocclusions may be carried out at the same time.

Treatment of RO is controversial and no consensus has yet been reached. The dentist should take into consideration factors such as the age of the patient, the medical history, previous dental experience, the number of affected teeth, the presence or absence of any pathology, as well as the attitude and wishes of the child and parent. The aims of treatment should include aiding mastication and speech, improving aesthetics, reducing the psychological impact, allowing normal jaw growth and development, and if possible, protection of any erupted affected teeth.

We present a case of RO that affected the primary and permanent dentitions in the anterior mandibular region, crossing the mid-line.

Case report

An 8.5-year-old girl was referred by her general dental practitioner to the Department of Paediatric Dentistry, Jordan University Hospital, Amman, Jordan, because of missing lower permanent incisors. The patient had neither other local abnormalities nor any relevant medical history. According to her parents, there had been a delay in the eruption of the lower primary incisors and canines, which had been grossly abnormal, with altered morphology, and yellowish/brownish in colour. The parents had not sought dental care for the child at that time and the lower anterior primary teeth had been lost very early after their eruption. There had been no similar cases among other members of the family.

On examination, the patient had no facial asymmetry or other extra-oral abnormality. In the maxillary arch, there were partially erupted permanent central incisors, primary lateral incisors, canines, and first and second molars in addition to the first permanent molars. In the mandibular arch, only the first and second primary molars and the first permanent molars were present; these teeth were clinically normal. The lower incisors and canines were absent, the associated alveolar mucosa was enlarged and covered by fibrous tissue (Fig. 1). The oral hygiene was poor, but there were no active carious lesions.

Panoramic, occlusal and peri-apical radiographs were taken. The maxillary dentition was normal, as were the mandibular primary molars, premolars and



Fig. 1. Intra-oral view of the lower arch showing unerupted permanent incisors and canines, and gingival enlargement.

permanent molars. In the anterior mandibular region, permanent incisors and canines were unerupted and showed retarded development for the age of the child. The teeth had thin radio-opaque contours with no distinction between enamel and dentine, 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. No, or only an insignificant amount of root formation, was visible radiographically (Fig. 2a,b).

On the basis of the clinical and the radiographic findings, a provisional diagnosis of RO was made.

Under local anaesthesia and intravenous sedation, all the unerupted mandibular incisors were enucleated, but the unerupted mandibular canines were retained. The enucleated teeth were of altered morphology, yellowish in colour, soft or rubbery in consistency, and had very short or unformed roots, and wide open apices.

Following decalcification, the extracted teeth were prepared for microscopic examination in the usual manner and stained with haematoxylin and eosin. Light microscopic examination showed dental hard tissue and associated fragments of dental follicles. The dental follicles were hyperplastic, and were composed of dense fibrous connective tissue containing odontogenic epithelial rests and whorled fibrous tissue. These were scattered foci of calcification within both the odontogenic epithelial rests and the whorled fibrous tissue (Fig. 3). The quantity of enamel matrix remaining after decalcification was inadequate for evaluation. The dentine was generally thin (Fig. 4) and contained a reduced number of dentinal tubules with irregular courses. Large amounts

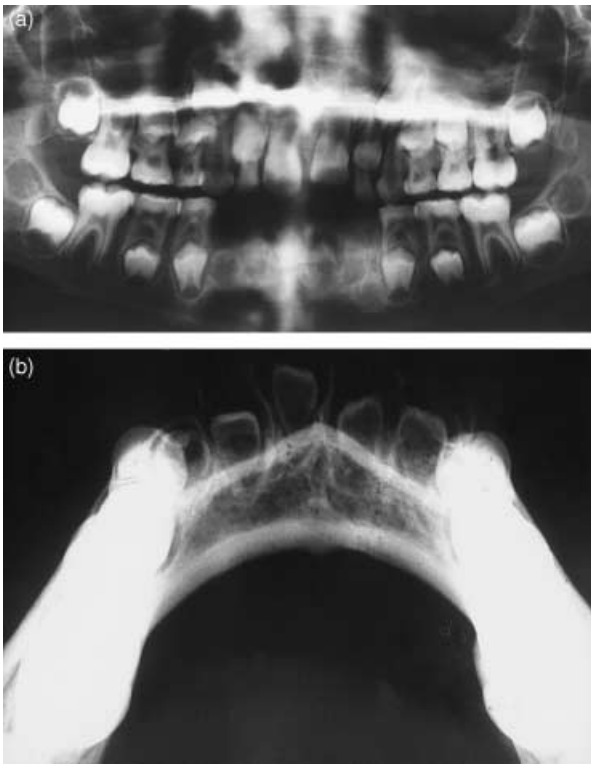


Fig. 2. (a) Panoramic tomogram showing normal maxillary dentition and the ghost-like appearance of the mandibular incisors and canines. (b) Occlusal view.

of interglobular dentine and globular masses interrupting the dentinal tubules were evident. Closer to the dentino-enamel junction, the dentine was more evenly calcified. Generally, the predentine zone was wider than that seen in normal teeth (Fig. 4). The roots were short and thin-walled with noticeable interglobular dentine and wide open apices (Fig. 5). The pulp chambers were large and contained focal areas of calcifications or stones. The odontoblasts were flattened and lying along the margin of predentine. The pulp tissue was intact and no necrosis was seen. Based on the histological features, the provisional diagnosis of RO was confirmed.

Two weeks postoperatively, a temporary acrylic mandibular partial denture was made to preserve the alveolar ridge during the period of skeletal growth (Fig. 6). Oral hygiene instructions, and dietary analysis and advice were given, and regular fluoride application was planned in order to prevent caries and periodontal disease. The patient was placed on periodic recall to review the unerupted mandibular permanent canines and to monitor the development of the mandibular arch.

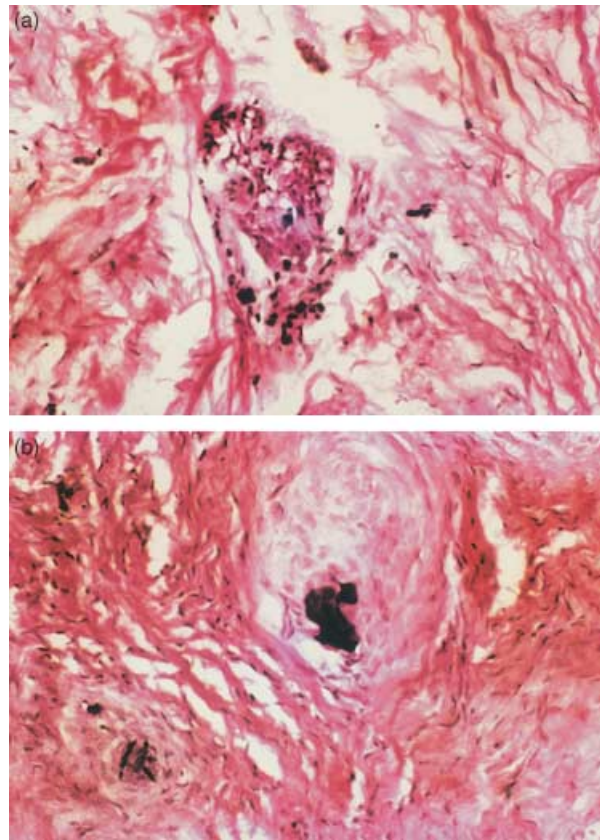


Fig. 3. (a & b) Calcifications within whorled fibrous tissue in the dental follicle (H & E, $\times 200$).



Fig. 4. Demineralized section showing the thin coronal dentine and a wide predentine zone (H&E, $\times 100$).

Discussion

To the best of our knowledge, the present case is the first case of RO to be reported in Jordan. The case is rare since the majority of the reported cases of RO affect one quadrant of the maxilla [12]. It is also rare for RO to cross the mid-line, as in our case;

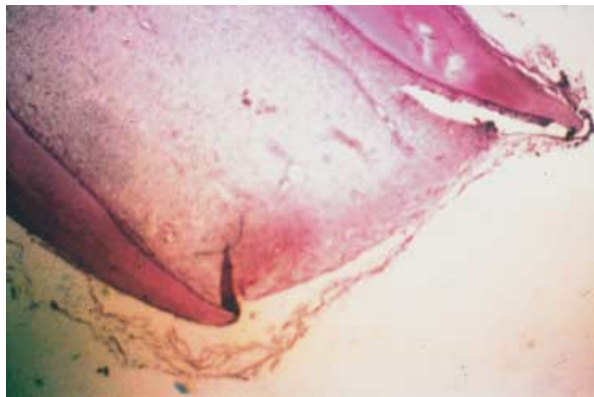


Fig. 5. Demineralized section showing the short and thin-walled root of an affected tooth with a wide open apex (H & E, $\times 100$).



Fig. 6. Clinical view taken after fitting the removable prosthesis replacing the lower incisors and canines.

this feature has been reported in only a small number of other studies [3,11,21,24]. The patient in this report had a history of deformed and brownish primary anterior teeth which were lost early in life, suggesting that the disturbance affected both dentitions in the same region. Although the precise aetiological 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.

The case showed features which were in accordance with the literature, in that the condition seems to affect females more than males, and most patients come to the dentist complaining of delayed eruption or gingival swelling [11,24,29]. Although these have been common, neither pain nor development of abscesses were reported by the child or her parents in this instance. Although some authors have

recently reported an association between RO and other abnormalities like ectodermal dysplasia, haemangiomas, hypophosphatesia, hydrocephalus and others [2,12,19,37,40–43], no such an association was found in the present case.

Since the clinical, radiographic and histological features of RO are so characteristic, dentists should face no difficulties in the diagnosis of this abnormality. The greater difficulty arises in treatment planning and no consensus as to the best option has yet been reached. The main question is whether to remove the affected teeth or not, but we believe that this decision should be taken after the assessment of each individual case of RO. Some authors prefer extraction and replacement with removable, and later, fixed prostheses [3,5,10,13,44,45], while others have suggested keeping the unerupted teeth, and waiting for their calcification and hoped for eruption [1,3,13,36]. When the affected unerupted teeth are retained, some authors have suggested that a removable temporary prosthesis may be constructed over them during the period of skeletal growth in order to preserve the alveolar ridge [12,51]. However, earlier reports have shown that delay in, or total failure of, eruption is common in RO [15,25,50]. Compared with the corresponding upper teeth, the calcification of the lower incisors and canines in this report was extremely delayed, and the time spent waiting for their calcification and eruption is likely to be prolonged. Even if they erupt, it is probable that they will have altered morphology and colour, as did their precursors, and the development of pathology, such as abscesses, may not be avoided. Gerlach *et al.* [24] have reported pulp necrosis in unerupted teeth afflicted by RO, complicating further any restorative treatment for these teeth if they do finally erupt. In treatment planning for RO, the wishes of the child and parents must also be taken into consideration. The parents of the child in this report were anxious about the fact that, if the teeth erupted, they would be discoloured and abnormal in shape in the same way as their predecessors. They were concerned that this would result in poor aesthetics and would cause unnecessary distress to their child. The affected teeth in our patient were surrounded by enlarged dental follicles which made their removal easy and minimally traumatic to the alveolus. Taking all these factors into consideration, the decision was made to remove the unerupted teeth and to replace them with a temporary acrylic prosthesis. The patient will be kept under review until

she reaches adulthood, when a final rehabilitation for loss of the incisors may be accomplished after the facial bones have stopped growing.

The permanent canines were also affected, but it was decided to leave these *in situ* in hopeful anticipation of their further calcification and eruption. This option was chosen because their location within the alveolus was deep and surgery to remove them might have endangered the adjacent, normally developing premolars. It has been reported that the maximum transverse growth of the mandible is attained at the time of eruption of the canines at around the age of 9 years [52]. Because our patient was 8.5 years old, extraction of the canines at this time might have affected this dimension of growth of the mandible. The canines may need extraction in the future, and final rehabilitation with implants and/or fixed prosthesis may prove necessary.

The care and treatment of a child with RO requires a multidisciplinary approach. Consultations between paediatric, prosthodontic and orthodontic specialties are necessary in each case of RO. Treatment planning should be designed for each individual case of RO, taking into account factors such as the age of the patient, the medical history, the extent of involvement, the eruption of the teeth, aesthetics, the development of pathology, and the wishes of the patient and parents. Regional odontodysplasia is not an aggressive lesion and simple extraction of the affected teeth is not followed by the development of any pathological lesions. There is, therefore, no need for resection of the teeth and/or the surrounding bone, as has been described in some reports [53,54]. Simple extraction has led to regression of the gingival enlargement associated with the affected teeth [13]. If the teeth afflicted by RO erupt and can function in the arch, or can be used for retention of prosthesis, then they should be retained wherever possible since such teeth have survived in some cases for more than 25 years [15].

Résumé. Les auteurs présentent un cas d'odontodysplasie régionale ou «dents fantômes», anomalie rare du développement, chez une Jordanienne de 8 ans $1/2$. L'anomalie touchait à la fois les incisives et canines temporaires et permanentes mandibulaires de façon bilatérale. Les dents maxillaires n'étaient pas affectées. C'est le premier cas décrit en Jordanie. Les caractéristiques cliniques, radiographiques et histologiques sont passées en revue. La prise en charge des patients atteints est discutée.

Zusammenfassung. Vorgestellt wird ein Fall der seltenen regionalen Odontodysplasie bei einem 8 jährigen jordanischen Mädchen. Die Anomalie betraf beidseits im Unterkiefer Schneidezähne und Eckzähne beider Dentitionen, Oberkieferzähne waren nicht betroffen. Es handelt sich um den ersten Fallbericht aus Jordanien. Klinische, röntgenologische und histologische Befunde werden abgehandelt. Die Therapie betroffener Patienten wird diskutiert.

Resumen. Los autores presentan un caso raro de anomalía del desarrollo denominada Odontodisplasia regional o “diente fantasma” en una niña jordana de 8, 5 años de edad. La anomalía afectaba tanto a incisivos inferiores como caninos temporales y permanentes, bilateralmente. Los dientes superiores no estaban afectados. Este es el primer caso de esta anomalía comunicada en Jordania. Se revisan los hallazgos clínicos, radiográficos e histológicos. Se discute el tratamiento de los pacientes afectados.

References

- 1 Marques AC, Castro WH, do Carmo MA. Regional odontodysplasia: an unusual case with a conservative approach. *British Dental Journal* 1999; **186**: 522–524.
- 2 Fanibunda KB, Soames JV. Odontodysplasia, gingival manifestations, and accompanying abnormalities. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontics* 1996; **81**: 84–88.
- 3 Lowry L, Welbury RR, Soames JV. An unusual case of regional odontodysplasia. *International Journal of Paediatric Dentistry* 1992; **2**: 171–176.
- 4 Cabral LA, Carvalho YR, Moraes E, Nogueira Tde O, Cavalcante AS, de Moraes LC. Regional odontodysplasia: a report of three cases. *Quintessence International* 1994; **25**: 141–145.
- 5 Hanks PA, Williams B. Odontodysplasia: report of two cases. *Pediatric Dentistry* 1998; **20**: 199–203.
- 6 Hitchin AD. Unerupted deciduous teeth in a youth aged 15 $1/2$. *British Dental Journal* 1934; **56**: 631–633.
- 7 McCall JO, Wald SS. *Clinical Dental Roentgenography*, 3rd edn. Philadelphia, PA: W. B. Saunders, 1947.
- 8 Rushton MA. A new form of dental dysplasia. *Oral Surgery* 1954; **7**: 543–549.
- 9 Zegarelli E, Kutscher A, Applebaum E, Archard A. Odontodysplasia. *Oral Surgery, Oral Medicine, Oral Pathology* 1963; **16**: 187–193.
- 10 Kahn MA, Hinson RL. Regional odontodysplasia. Case report with etiologic and treatment considerations. *Oral Surgery, Oral Medicine, Oral Pathology* 1991; **72**: 462–467.
- 11 Lustmann J, Klein H, Ulmanky M. Odontodysplasia. Report of two cases and review of the literature. *Oral Surgery, Oral Medicine, Oral Pathology* 1975; **39**: 781–793.
- 12 Crawford PJ, Aldred MJ. Regional odontodysplasia: a bibliography. *Journal of Oral Pathology and Medicine* 1989; **18**: 251–263.
- 13 Gomes MP, Modesto A, Cardoso AS, Hespanhol W. Regional odontodysplasia: report of a case involving two separate

- affected areas. *ASDC Journal of Dentistry for Children* 1999; **66**: 203–207.
- 14 Pullon PA, Miller AS. Ghostlike tooth. *General Dentistry* 1984; **32**: 526.
 - 15 Gibbard PD, Lee KW, Winter GB. Odontodysplasia. *British Dental Journal* 1973; **135**: 525–532.
 - 16 Ansari G, Reid JS, Fung DE, Creanor SL. Regional odontodysplasia: report of four cases. *International Journal of Paediatric Dentistry* 1997; **7**: 107–113.
 - 17 Fearnie J, Williams DM, Brook AH. Regional odontodysplasia: a clinical and histological evaluation. *Journal of the International Association of Dentistry for Children* 1986; **17**: 21–25.
 - 18 Hovinga J, Ingenhous R. Regional odontodysplasia. *International Journal of Oral Surgery* 1979; **8**: 474–477.
 - 19 Steiman HR, Cullen CL, Geist JR. Bilateral mandibular regional odontodysplasia with vascular nevus. *Pediatric Dentistry* 1991; **13**: 303–306.
 - 20 Walton JL, Witkop CJ, Jr, Walker PO. Odontodysplasia. Report of three cases with vascular nevi overlying the adjacent skin of the face. *Oral Surgery, Oral Medicine, Oral Pathology* 1978; **46**: 676–684.
 - 21 Galeone RJ, Phillips JF, Pincok DG. Odontodysplasia. *Oral Surgery, Oral Medicine, Oral Pathology* 1970; **29**: 879–880.
 - 22 Reade PC, Radden BG, Barke JJ. Regional odontodysplasia. A review and a report of two cases. *Australian Dental Journal* 1974; **19**: 152–161.
 - 23 Hankey GT, Duckworth R. Odontodysplasia in the deciduous dentition. *Dental Practitioner and Dental Record* 1968; **19**: 93–95.
 - 24 Gerlach RF, Jorge J, Jr, de Almeida OP, Colletta RD, Zaia AA. Regional odontodysplasia. Report of two cases. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontics* 1998; **85**: 308–313.
 - 25 Sabah E, Eden E, Unal T. Odontodysplasia: report of a case. *Journal of Clinical Pediatric Dentistry* 1992; **16**: 115–118.
 - 26 Ferguson FS, Creath CJ, Buono B. Infraorbital infection related to Odontodysplasia. Case report. *Pediatric Dentistry* 1990; **12**: 397–400.
 - 27 Anneroth G, Ramstrom G. Unilateral odontodysplasia. *Swedish Dental Journal* 1980; **4**: 93–100.
 - 28 Rushton MA. Odontodysplasia: ghost teeth. *British Dental Journal* 1965; **119**: 10–13.
 - 29 Gardner DG, Sapp JP. Regional odontodysplasia. *Oral Surgery* 1973; **35**: 351–365.
 - 30 Hintz CS, Peters RA. Odontodysplasia. Report of an unusual case and review of the literature. *Oral Surgery, Oral Medicine, Oral Pathology* 1972; **34**: 744–750.
 - 31 Bergman G, Lysell L, Pindborg JJ. Unilateral dental malformation: report of two cases. *Oral Surgery* 1963; **16**: 48–60.
 - 32 Gardner DG. The dentinal changes in regional odontodysplasia. *Oral Surgery, Oral Medicine, Oral Pathology* 1974; **38**: 887–897.
 - 33 Gould AR, Farman AG, Marks ID. Pericoronal features of regional odontodysplasia. *Journal of Oral Medicine* 1984; **39**: 236–242.
 - 34 Kerebel LM, Kerebel B. Soft-tissue calcifications of dental follicle in regional odontodysplasia: a structural and ultrastructural study. *Oral Surgery, Oral Medicine, Oral Pathology* 1983; **56**: 396–404.
 - 35 Kinirons MJ, O'Brien FV, Gregg TA. Regional odontodysplasia: an evaluation of three cases based on clinical, microradiographic and histopathological findings. *British Dental Journal* 1988; **165**: 136–139.
 - 36 Vaikuntam J, Tatum NB, McGuff HS. Regional odontodysplasia: review of the literature and report of a case. *Journal of Clinical Pediatric Dentistry* 1996; **21**: 35–40.
 - 37 Schmid-Meier E. Unilateral odontodysplasia with ipsilateral hypoplasia of the mid-face. A case report. *Journal of Maxillofacial Surgery* 1982; **10**: 119–122.
 - 38 Sadeghi EM, Ashrafi MH. Regional odontodysplasia: clinical, pathologic, and therapeutic considerations. *Journal of the American Dental Association* 1981; **102**: 336–339.
 - 39 Melamed Y, Harnik J, Becker A, Shapira J. Conservative multidisciplinary treatment approach in an unusual odontodysplasia. *ASDC Journal of Dentistry for Children* 1994; **61**: 119–124.
 - 40 Pandis N, Polido C, Bell WH. Regional odontodysplasia. A case associated with asymmetric maxillary and mandibular development. *Oral Surgery, Oral Medicine, Oral Pathology* 1991; **72**: 492–496.
 - 41 Dahllof G, Lindskog S, Theorell K, Ussiso R. Concomitant regional odontodysplasia and hydrocephalus. *Oral Surgery, Oral Medicine, Oral Pathology* 1987; **63**: 354–357.
 - 42 Slootweg PJ, Meuwissen PR. Regional odontodysplasia in epidermal nevus syndrome. *Journal of Oral Pathology* 1985; **14**: 256–262.
 - 43 Russell K, Yacobi R. Generalized odontodysplasia concomitant with mild hypophosphatasia – a case report. *Journal of the Canadian Dental Association* 1993; **59**: 187–190.
 - 44 Ferguson JW, Geary CP. Regional odontodysplasia. *Australian Dental Journal* 1980; **25**: 148–151.
 - 45 O'Neil DW, Koch MG, Lowe JW. Regional odontodysplasia: report of case. *ASDC Journal of Dentistry for Children* 1990; **57**: 459–461.
 - 46 Guckes AD, McCarthy GR, Brahim J. Use of endosseous implants in a 3-year-old child with ectodermal dysplasia: case report and 5-year follow-up. *Pediatric Dentistry* 1997; **19**: 282–285.
 - 47 Bergendal T, Eckerdal O, Hallonsten AL, Koch G, Kurol J, Kvint S. Osseointegrated implants in the oral habilitation of a boy with ectodermal dysplasia: a case report. *International Dental Journal* 1991; **41**: 149–156.
 - 48 Becktor KB, Becktor JP, Keller EE. Growth analysis of a patient with ectodermal dysplasia treated with endosseous implants: a case report. *International Journal of Oral and Maxillofacial Implants* 2001; **16**: 864–874.
 - 49 Lowe O, Duperon DF. Generalized odontodysplasia. *Journal of Pedodontics* 1985; **9**: 232–243.
 - 50 Pinkham JR, Burkes EJ, Jr. Odontodysplasia. *Oral Surgery, Oral Medicine, Oral Pathology* 1973; **36**: 841–850.
 - 51 Masuda N, Kitamura K, Ootsuki K, Sobue S, Furukawa S, Fuchihata H. Regional odontodysplasia: report of a case. *Osaka Daigaku Shigaku Zasshi* 1983; **28**: 287–294.
 - 52 Foster TD, Grundy MC. Occlusal changes from primary to permanent dentitions. *British Journal of Orthodontics* 1986; **13**: 187–193.
 - 53 Miller WA, Seymour RH. Odontodysplasia. *British Dental Journal* 1968; **125**: 56–59.
 - 54 Werelds RJ, Douniau R. Ghost teeth (odontodysplasia, an unusual anomaly). *Acta Stomatologica Belgica* 1972; **69**: 375–427.

Copyright of International Journal of Paediatric Dentistry is the property of Blackwell Publishing Limited and its content may not be copied or emailed to multiple sites or posted to a listserv without the copyright holder's express written permission. However, users may print, download, or email articles for individual use.