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Regional odontodysplasia involving maxillary right quadrant treated by dental implant prosthetic rehabilitation: A review and case report

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Abstract
Regional odontodysplasia (ROD) is a rare dental anomaly that can affect deciduous and permanent dentition. This unique dental abnormality involves enamel, dentin, pulp, and dental follicle. ROD has a prevalence of less than 1:1.000.000 and the age at the time of diagnosis ranges from 1 to 23 years. Females are slightly more affected than males at a ratio of 1.4:1. The maxilla is affected twice as often as the mandible. The aetiology of ROD is still unknown and conditions such as viral infections, local trauma, vascular defects, irradiation, metabolic disturbance, rhesus incompatibility and medications during pregnancy have been suggested as possible causes.
The diagnosis is usually made by clinical and radiographic findings. Generally, the affected teeth fail to erupt, but when erupted, they are small, hypoplastic, or hypocalcified with deep pits and clefts which could be connecting the pulp. Radiographically, the affected teeth show a typical “ghost-like” appearance. The affected teeth have large pulp chambers and short roots with open apices. The treatment of ROD is controversial. It is widely accepted that implant-supported single-unit crowns represent a reliable treatment option for the replacement of missing teeth with favourable outcomes.
The objective of this review and reported case highlights epidemiology, etiopathogenesis, diagnosis, and implant treatment modality.

Keywords: Regional odontodysplasia, epidemiology, etiopathogenesis, diagnosis, treatment

Introduction
ROD is a rare dental anomaly of unknown cause that affects both deciduous and permanent teeth [1]. ROD is also known as odontogenesis imperfecta, odontogenic dysplasia and non-hereditary amelogenesis imperfecta, and ghost teeth, in which both ectodermal- and mesodermal-derived tooth tissues are affected [2]. This condition was probably first described by Hitchin (1934) [3]. The first review of this condition was published by McCall and Wald in 1947, but the term "odontodysplasia" was presented by Zegarelli *et al* in 1963 [4]. In the late 1970s, Pindborg added the term 'regional' to emphasize the local nature of this condition [5]. The ROD presents clinical, radiological, and histopathological features that support the diagnosis [6]. This unique dental anomaly affects the enamel, dentin, pulp, and dental follicle and generally disturbs teeth in one quadrant [7]. Other disorders that share some features with ROD include dentine dysplasia types I and II, shell teeth, rickets, hypophosphatasia and amelogenesis imperfecta. However, it should be noted that, unlike ROD, all of these conditions affect the entire dentition, in contrast to the partial involvement seen in ROD [8]. Occasionally, ROD is associated with other conditions such as vascular nevi, affected hemifacial malformations, hydrocephalus, and hypophosphatasia [9].
Developmental dental abnormalities can lead to esthetic and functional dilemmas if left untreated. Implant-supported single crowns are generally accepted as a reliable treatment option for the replacement of missing teeth with perfect treatment outcomes in terms of high implant survival and steady peri-implant marginal bone ranges [10].

Epidemiology

The prevalence of this condition is not entirely clear, epidemiologically the study was mostly case report-based [11]. The prevalence of ROD is less than 1:1,000,000 and approximately 168 cases have been published in English by 2019 [6]. Age at diagnosis ranges from 1 to 23 years [12]. Females are slightly more affected than males in a ratio of 1.4:1 [13]. ROD is usually unilateral and does not tend to cross the midline [14]. The maxilla is affected twice as often as the mandible, with the left quadrant of the maxilla most commonly affected [15]. Central and lateral incisors are affected more frequently than posterior teeth [3].

Etiopathogenesis

The cause of ROD is obscure and conditions such as viral infections, local trauma, vascular abnormalities, radiation, metabolic disorders, rhesus incompatibility, and medications during pregnancy are thought to be the possible causes [16]. According to research done by Crouson *et al*., there is an increase in the levels of matrix metalloproteinases (MMPs) which are essential during tooth formation and also an increase in their natural inhibitors (TIMP- 1 and -2) in ROD. However, the parallel increase in natural inhibitors was not sufficient to inhibit the enzymatic activity of MMP, leading to tissue destruction. The mechanism involved in this increase in enzymes remains unclear [17]. The distribution of affected teeth in the four quadrants led some authors to suggest that somatic mutations may be a causative factor [5].

Diagnosis and characteristics

Diagnosis is usually based on clinical and radiographic findings, sometimes supplemented by histopathological examination [2]. Clinically, swellings, gingivitis, and abscesses are correlated to both periapical and periodontal infections [9]. Typically, the afflicted tooth may not erupt when the affected tooth erupts, it appears small, hypoplastic, or hypocalcified with deep depressions and fissures where the pulp can connect [18].

Radiographically affected teeth have a typical “ghost” appearance [19]. This condition is also referred to as a “shell tooth” to describe radiographic features such as hypoplasia and hypocalcification of enamel and dentin [20]. Affected teeth have large pulp chambers, short roots, and open apices [21]. Histological features include hypocalcified dental hard tissues and areas of atypical and degenerative hypocalcification [22]. However, in the ground section, the enamel is uneven in thickness and has a laminated appearance. Dentin contains fissures scattered through a mixture of spherical dentin and amorphous material as well as a globular area of disorganized tubular dentin [23].

Management

Treatment of ROD is argumentative and requires a multidisciplinary approach.

The main problem is whether the affected tooth should be extracted [28]. According to the literature, the most common cause of tooth extraction is abscess formation [24]. The main goals of treatment are space maintenance, restoration of masticatory function, esthetics consideration, and reduction of the psychological consequences of tooth loss. As a result, consultation between pediatric, orthodontic, orthopaedic and surgical specialists is often necessary [14]. Autologous transplantation of healthy teeth has been proposed even in the presence of normal donor teeth [25]. The patient's age and cooperation, dental prognosis, and follow-up examinations have a significant impact on elective treatment (11). As reported in the literature, implantation is a productive, profitable, and predictable option for restoring missing teeth [26].

Case report

A 20 years old Libyan female patient was referred to Dar Alfardous dental clinic, with the chief complaint of an esthetic and functional problem in the upper right jaw. There was no relevant medical or family history of any systemic disease.

Upon extra oral examination, no facial abnormality was detected. The skin was normal with non-palpable regional lymph nodes.

The Federation Dentaire Internationale (FDI) tooth numbering system was used to record the related teeth in the affected region of the jaw.

Parents gave a history of previous teeth extraction after the complete eruption of the deciduous teeth because of the presence of small brown discoloured abnormal teeth and gingival swelling with redness.

On intraoral examination, the patient was wearing a removable partial denture. There was no carious lesion, with normal occlusion, soft tissues and developing dentition except for the maxillary right quadrant. On the right side of the maxilla, all the teeth were missing, with no gingival swelling or redness. Fig. 1

The previous digital panoramic radiograph revealed obvious severe morphological alteration and a very thin tooth structure with a faint outline appearance due to marked reduced radiodensity of enamel and dentine structures which affected the teeth from 12 to 15 on the affected side while 17and18 teeth were impacted and 16 was missing. Furthermore, the affected teeth were devoid of root development and no distinction between enamel and dentin structures. Fig.2. The dentition in the other quadrants was normal. According to the past dental history, previous radiographic picture and clinical criteria, the diagnosis of regional odontodysplasia was given. Prosthetic rehabilitation with a dental implant for the missing teeth was performed instead of a removable partial denture. Fig. 3.

Discussion

ROD is a rare developmental anomaly that tends to be localized and affects the ectodermal and mesodermal components of the tooth [13]. ROD is more common in women without a racial tendency [4]. Our patient was a young adult woman. It was consistent with much previous literature regarding age and gender at the time of diagnosis.

The upper jaw is affected twice as often as the lower jaw. It is usually unilateral and rarely crosses the midline [8]. In the presented case morphological alteration of affected teeth 11 to 15 with complete impacted 17 and 18 at the right maxillary quadrant. This finding was in agreement with Alotaibi 2019 [12] who found maxillary predilection by analyzing 161 cases of ROD.

The diagnosis of ROD depends on clinical and radiographic findings [11]. In our patient, the most frequent clinical symptom was that all the teeth in the right maxillary quadrant were missing. Moreover, the radiographic criteria involving the permanent dentition in the maxillary right quadrant such as obvious severe morphological alteration and very thin tooth structure with faint outline appearance due to marked reduced radiodensity of enamel and dentine structures emphatically upheld the definite diagnosis of the presented condition. These characteristics coincided with the most reported cases

previously.

Although dental implants are the preferred treatment option to replace missing teeth, the success of implants depends on the precision and accuracy of implant placement for optimal prosthetic results [27]. In our case, a dental implant was performed as the treatment of choice for replacing the missing teeth resulting in the successful restoration of masticatory function and appropriate esthetic outcomes.

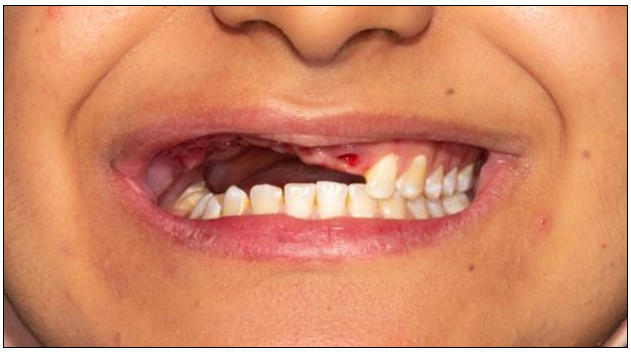

Fig 1: Photograph of the case showing missing teeth in right maxillary quadrant

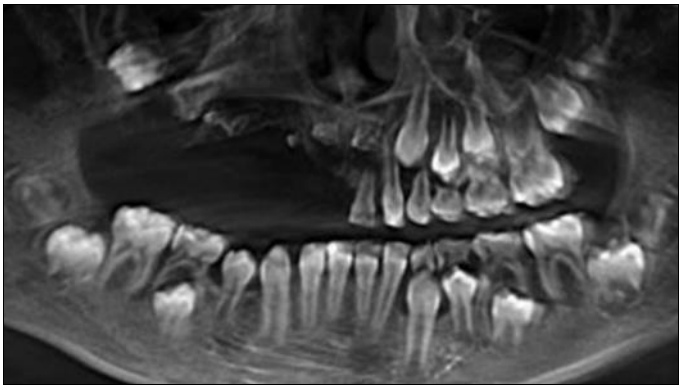

Fig 2: Previous digital panoramic radiograph was taken in childhood stage. The affected teeth revealed a faint outline appearance (red arrow), devoid of root development and no distinction between enamel and dentin structures (yellow arrow)

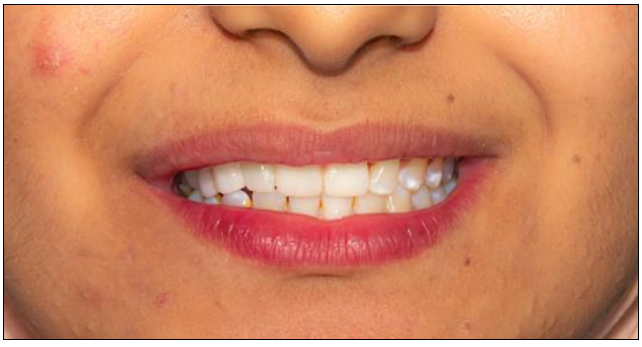

Fig 3: Photograph of the case showing esthetic and masticatory efficiency restored by a dental Implant.

Conclusion

ROD is a rare odontogenic developmental abnormality of uclear aetiology with anomalies of dentin, enamel and cementum. Both dentitions can be affected.

When the clinical pathognomic signs diminish as a result of extraction or surgical intervention, the previous radiography image and past dental history are highly useful diagnostic tools.

The main goals of ROD treatment are to improve aesthetics, restore masticatory function, and reduce psychological effects. When treating young adults who have missing teeth, the dental implant can be considered as a promising therapeutic strategy.

A more detailed study of ROD is needed to identify possible etiological factors.

Conflict of Interest

Not available

Financial Support

Not available

References

1. Cunha J, *et al*. Regional Odontodysplasia Affecting the Maxilla. Head and Neck Pathology. 2020;14:224-229.
2. Mukhopadhyay S, Roy P, Halder M. Regional odontodysplasia in the primary dentition associated with eruption failure. Int J Health Allied Sci. 2016;5:195-7.
3. Magalhães AC, *et al*. Regional odontodysplasia: case report. J Appl Oral Sci. 2007;15(6):465-9.
4. Mathew A, *et al*. Ghost teeth: Regional odontodysplasia of maxillary first molar associated with eruption disorders in a 10-year-old girl. J Pharm Bioall Sci. 2015;7:800-3.
5. Ohoud Nasser Alotaibi. Regional odontodysplasia: an overeview. Paki Oral & Dent J. 2015;35(2):175-178.
6. Riolobos González MF, *et al*. Clinical, Radiographic and Histologic Evaluation of Regional Odontodysplasia*:* an Unusual Case Report with 6-Year Follow-Up. Int J Dent Oral Health. 2020;7(2):1-6.
7. Koruyucu M, *et al*. Management of regional odontodysplasia: a 10-year-follow-up case report and literature review. Eur Oral Res. 2018;52(2):111-6.
8. Mehta DN, Bailoor D, Patel B. Regional odontodysplasia. J Indian Soc Pedod Prev Dent. 2011;29:323-6.
9. Koskinen S. Familial oligodontia and regional odontodysplasia associated with a PAX9 initiation codon mutation. Clin Oral Invest. 2019;23:4107-4111.
10. Roccuzzo A, *et al*. Narrow diameter implants to replace congenital missing maxillary lateral incisors: A 1-year prospective, controlled, clinical study. Clin Oral Impl Res. 2022;33:844-857.
11. Jahanimoghadam F, Pishbin L, Rad M. Clinical, Radiographic, and Histologic Evaluation of Regional Odontodysplasia: a Case Report with 5-year Follow-up. J Dent Shiraz Univ Med Sci. 2016;17(2):159-163.
12. Alotaibi O, Alotaibi G, Alfawaz N. Regional odontodysplasia: An analysis of 161 cases from 1953 to 2017. SDJ. 2019;31:306–310.
13. Barberia E, *et al*. Regional odontodysplasia. A literature Review and three case reports. Euro J Paed Dent. 2012;13(2):161-166.
14. Babu N, Smriti RJ, Pratima DB. Regional odontodysplasia: Report of an unusual case involving mandibular arch. Contemp Clin Dent. 2015;6:237-9.
15. Vinod Upadhyay, *et al*. Regional Odontodysplasia Crossing Midline. A Rare Case Report. IJCPD. 2011;4(2):159-161.
16. Ramakrishnan M, Menon P. Odontodysplasia involving single tooth: A rare entity. J Res Dent Sci. 2014;5(2):140-142.
17. Murthy PS, Deshmukh S. Regional odontodysplasia and its treatment maneuver. Int J Health Allied Sci. 2013;2:153-8.

18. Tervonen SA, *et al*. Regional odontodysplasia: a review of the literature and report of four cases. Clin Oral Invest. 2004;8:45–51.
19. Desai VD, *et al*. Regional odontodysplasia with supernumerary teeth in pediatric patients: Coincident new finding? Chrisme J Health Res. 2015;2:272-5.
20. Sikdar R, *et al*. Regional odontodysplasia: A rare case series and review. J Prim Care Dent Oral Health. 2021;2:88-91.
21. Davi de Sá Cavalcante, *et al*. Mandibular Regional Odontodysplasia in an 8-year-old Boy showing Teeth Disorders, Gubernaculum Tracts, and Altered Bone Fractal Pattern. Int J Clin Pedi Dent. 2018;11(2):128-134.
22. Malhotra R, *et al*. Regional odontodysplasia: A classical case report. J Res Dent Sci. 2013;4:86-9.
23. Heera R, *et al*. Regional odontodysplasia of mandible-report of a rare case and literatue review. Int J Sci Res. 2018;7(7):48-49.
24. Bowden E, *et al*. Cervicofacial infection in a 3-year-old child with regional odontodysplasia: A case report and literature review. J Oral Maxillofac Pathol 2018; 22:S117-20.
25. Srivathsa SH. Regional odontodysplasia: A case with radiographic evidence of advancing development. Int J Orofac Res. 2018;3:37-9.
26. Mukhopadhyay S, Roy P, Halder M. Regional odontodysplasia in the primary dentition associated with eruption failure. Int J Health Allied Sci. 2016;5:195-7.
27. Rawal S. Guided innovations: Robot-assisted dental implant surgery. J Prosthet Dent. 2022;127(5):673-4.

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