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CASE REPORT

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Regional odontodysplasia diagnosed and treated *via* multidisciplinary approach: a case report

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Abstract

Regional odontodysplasia (RO) is a rare non-hereditary dental anomaly associated with dysplasia. Its etiology remains unclear but is known to affect both the mesodermal and ectodermal dental components, as well as deciduous and permanent dentitions. Its young age of onset and complexity has great physical and psychological impact on the affected patients. However, the clinical management of RO remains unified without standardized treatment guidelines. Thus, this study aimed to report an RO case, the first from Jiangxi Province, China, and discuss its clinical diagnosis and treatment to provide a reference to treat similar cases more effectively in the future.

Keywords

Odontodysplasia; Ghost teeth; Diagnosis; Therapy; Etiology

1. Introduction

Regional odontodysplasia (RO) refers to tooth developmental disorders and localized eruption abnormalities of the maxilla or mandible that may also involve deciduous or/and permanent teeth [1]. It is a rare anomaly, with fewer than 200 cas es reported until April 2022 [2, 3]. Its diagnosis is mainly ba sed on the patient's clinical presentation, imaging features and histopathological findings [2]. Currently, the clinical treat ment of RO remains unified without standardized treatment g uidelines. Although some researchers recommend that all aff ected teeth, except those causing abscesses, should be preser ved as much as possible, preferably until the permanent restor ation is performed [4], there are still controversies in its mana gement,especially regarding the tooth extraction, timing an d choice treatments [5]. The prognosis of these patients re mains un-known due to the early age of onset and comple xity of the disease. This article reports a case of RO that was s uccessfully managed by a multidisciplinary approach.

2. Case report

The patient was a boy aged 7 years and 10 months. On June 2017, he was brought by his mother to the Department of Oral and Maxillofacial Surgery of the Affiliated Stomatological Hospital of Nanchang University due to the complaint of recurrent swelling, discharge of pus from the left maxilla and delayed eruption of the maxillary left permanent tooth. His previous (prenatal and birth), medical and family history were unremarkable, and he denied a history of trauma. Extraoral examination showed symmetrical maxillofacial structures, while intraoral examination showed dark red gingiva and an obvious swelling at the vestibular sulcus of the teeth (#63, #64). The swelling was about 2 cm × 3 cm, soft, without pulsation and had a small amount of pus overflow on palpation. Mixed dentition with many teeth (#61, #62, #63, #65) was observed with residual roots.

In addition, residual roots of the maxillary left deciduous teeth were observed, as well as delayed and malformed tooth germ in permanent teeth with thin hard tissues, wide medullary cavity, short root, and low density that resembled ghost teeth (Fig. 1).

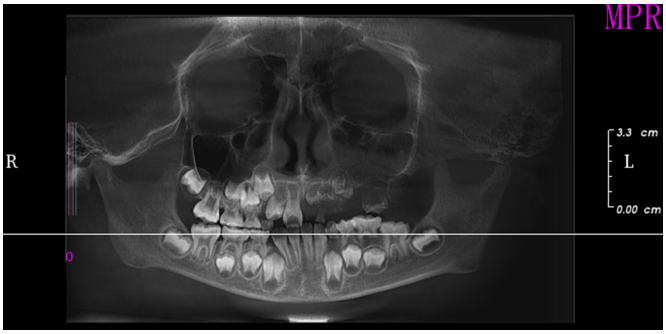

FIGURE 1. Cone Beam Computed Tomography (CBCT) film showing residual roots of the maxillary left deciduous teeth, with delayed and malformed tooth germ in permanent teeth with thin hard tissues, wide medullary cavity, short root, and low density.

Based on these features, the patient was diagnosed with RO, following which he underwent abscess resection and residual root (#61, #62, #63, #65) extraction by oral and maxillofacial surgeons under general anesthesia. Intraoperative frozen biopsy confirmed the presence of a left maxillary cyst with infection.

The maxillary left permanent incisors had not yet erupted by the time he was 8 years old and did not have redness or swelling in the mucosa. Follow-up radiographs showed delayed tooth germ development and blurred images. Mandibular and right maxillary development showed no abnormalities (Fig. 2).

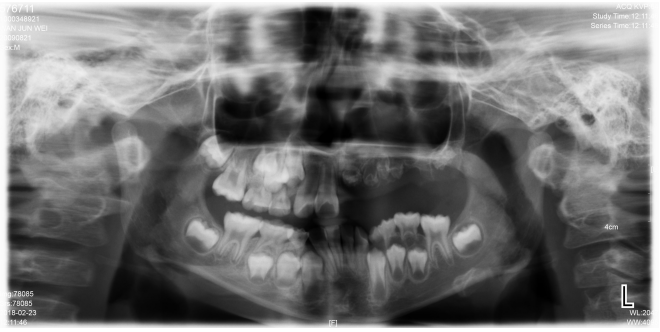

FIGURE 2. Panoramic radiograph showing unerupted permanent tooth germs in the upper left side, delayed tooth germs development, and blurred images. No abnormalities were observed in mandibular and right maxillary development.

The unerupted teeth germ demonstrated increased density by the time the boy was 9 years old, while teeth #25 and #27 had no germs (Fig. 3).

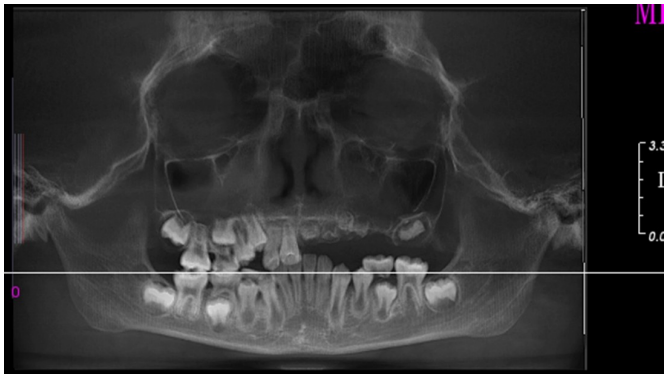

FIGURE 3. CBCT showing increase in image density of the unerupted teeth germ and no tooth germs for teeth #25 and #27.

In addition, there were no significant changes in the maxillary left permanent tooth germs (Fig. 4). With the patient's and parents' consent, transitional restorations with maxillary removable partial dentures were planned by a prosthodontist when the boy was 10 years old (Fig. 5). Then, when the patient was 11 years old, full-mouth deciduous tooth replacement was completed. However, the hard tissues of the tooth embryo remained thin, and the pulp chamber was wide, exhibiting a "ghost-like" appearance (Fig. 6).

The boy was advised for further treatment at the orthodontics department due to crowded dentition (Fig. 7), and the patient and family provided informed consent. The treatment plan was to refabricate the removable partial dentures after orthodontic treatment. Further, following subsequent routine follow-ups, the options for final rehabilitation would be selected according to the condition of impacted permanent tooth germs in adulthood.

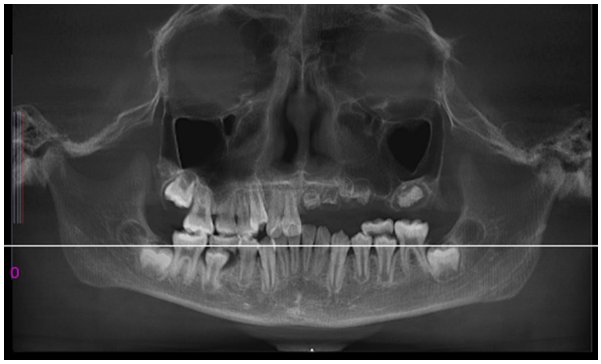

FIGURE 4. CBCT showing no significant changes in the left maxillary permanent tooth germs.

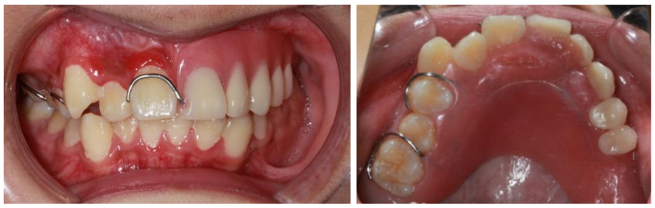

FIGURE 5. Buccal and occlusal view of the removable partial oral dentures.

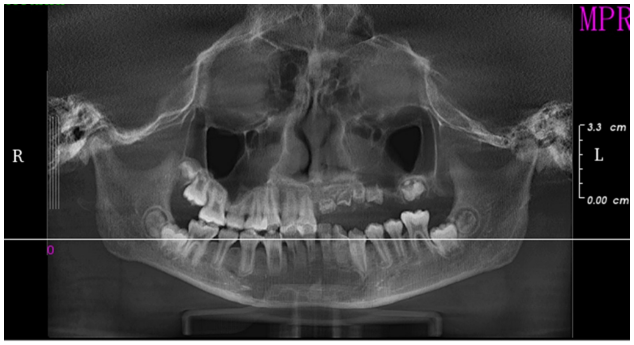

FIGURE 6. CBCT showing no significant development of the left maxillary permanent tooth embryo and a slight increase in density. However, the hard tissues remained thin, with a wide pulp chamber, demonstrating a "ghost-like" appearance.

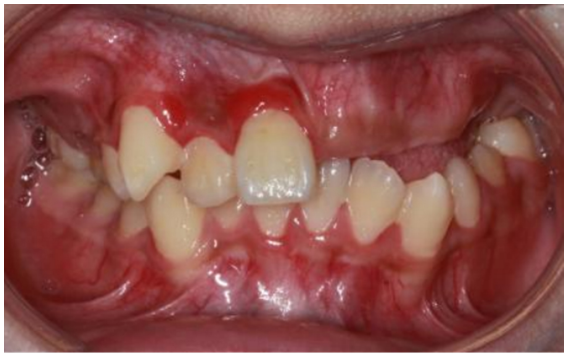

FIGURE 7. Illustration of the crowded dentition of the patient.

3. Discussion

3.1 Origin and etiology

RO was first described by Hitchin in 1934, following which many researchers started to report its associated symptoms and radiographic features [6–8]. Although its etiology remains unclear [3, 9], associations with local injury, infection, radiation, teratogenic drugs, high fever, malnutrition, local blood circulation disorders, genetic mutations and disturbances in neural crest cell migration have been proposed [10–14]. Previous literature showed the involvement of Sonic hedgehog (SHH) signaling in regulating root elongation [15, 16]. Additionally, mutations in Patched (PTC), the receptor for SHH, were shown to inhibit cell proliferation in the dental mesenchyme. PTC gene can lead to nevoid basal cell carcinoma syndrome in humans [17–19], indicating the SHH pathway as a potential mechanism to explain the pathogenesis of RO. Furthermore, mutations in the *Paired box (PAX) 9* gene were observed in patients with familial oligodontia and RO, and it is believed that tooth agenesis and RO may have a common etiology, that is, associated with mutations in the *PAX9* start codon [13]. However, further studies are required for confirmation.

3.2 Diagnosis and differential diagnosis

The diagnosis of RO is mostly based on clinical symptoms and radiological image features [2] and sometimes on histopathological examination. It often occurs in children and adolescents and with a recorded peak at ages 2–5 and 8–12 years. In most cases, both dentitions are involved, and the maxilla is more commonly affected [20]. The ratio of males to females is reported to be 1:1.37 [2]. RO often occurs in a single quadrant with several consecutive teeth, and the lesions are more common in the anterior part [21], but in rare occasions, multiple quadrants and the whole mouth might be affected [22, 23]. However, no racial or geographical variability has been reported so far [24].

The most common complaints of patients are abnormal tooth eruption, followed by gingiva swelling, which may sometimes even spread to check and neck infection [25]. The teeth involved have abnormal morphology with yellow or brown coloring, rough surface and low hardness on probing [10, 26]. Children often have malocclusion [23], and RO occurring in the anterior region may result in a shifted midline [4]. The imaging of the affected teeth shows wide pulp chambers with open apices and thin contours, revealing abnormal formation and mineralization [24]. There is an indistinct boundary between enamel and dentin, and the radiodensity may be lower than in normal teeth [9]. The morphology of these teeth is small and peculiar, thereby exhibiting a “ghost-like” appearance [3]. The main performance of pathological sections is a deficient and abnormal formation of dentin and enamel [5, 27]. Enamel prisms are usually irregular, the enamel can be hypoplastic and hypocalcified, and the dentin-enamel junction is irregular in some areas. The number of dentin tubules is reduced, showing a distorted morphology. Foci of calcification are visible in the pulp cavity to varying degrees [21]. Some researchers surmised that the structural disorganization of the affected teeth might result from an imbalance of matrix metalloproteinases (MMPs) and tissue inhibitors of matrix metalloproteinases (TIMPs) [28].

Clinical differentiation is required from other causes of tooth eruption abnormalities and genetic disorders [5, 26], such as dentinogenesis imperfecta (DI), primary failure of eruption (PFE) and cleidocranial dysplasia (CCD) [29, 30].

3.3 Treatment

RO patients are often young, with different areas and degrees of involvement and unknown prognosis of the affected teeth, making the disease presentation and treatment complex and diverse. Although doctors recommend multidisciplinary treatment, whether to extract the affected tooth or not remains controversial [31].

When the involved tooth becomes severely infected or loose, it cannot be restored. As shown in the case above, the boy suffered from an acute abscess, and the residual roots were extracted as soon as possible to control the infection. Extraction of the affected tooth followed by partial denture restorative treatment is considered the optimal treatment when: the affected tooth is extremely hypoplastic and lacks support; pulpal necrosis is complicated by abscess; presence of unilateral RO leading to midline deviation (removable partial denture can prevent aggravation); and tooth dysplasia causing severe psychological impact [4]. Patients who are young or have several impacted teeth involving multi-quadrants that need to be extracted can be considered for a single procedure under general anesthesia, which may minimize patient trauma [32, 33]. As vascular anomalies might be associated with RO, the patients should undergo systematic preoperative examination before any procedure is performed [34]. In addition, special attention should be paid to bleeding and other conditions during and after surgery [13].

Preserving the affected teeth as long as possible is recommended by many doctors because it can maintain good mastication and aesthetics for the child [35]. Nijakowski [3] recommended that the affected teeth should not be extracted until the inflammation has occurred and the surrounding bone has developed. The impacted teeth may continue to develop during follow-up observations [14, 36]. However, severe periapical infections and maxillofacial abscesses can occur later in all affected teeth [4], as well as some sequelae of retained epithelial tissue, such as the formation of follicular cysts or odontogenic tumors and/or pain induced by nervous pressure on the mandibular canal [37]. Thus, regular follow-up is very important for these patients to monitor the disease progression and provide timely treatments. Additional measures that can be taken to protect the teeth include fluoridization, pit and fissure sealant, crowning, and others [1].

As for incomplete eruption, orthodontic treatment and periodontal crown lengthening are advised [38]. However, teeth should have well-developed roots or demonstrate continued development during observation. As shown in the case above, the boy needed orthodontic treatment to line the dentition to provide enough space for future restoration.

Gap retainers can be fabricated for gap management after single tooth extraction [39], but restorative treatment should be made for patients with multiple extracted teeth or teeth

with abnormal formation [1, 24, 40, 41]. Prosthodontic treatment can maintain the patient's mastication and aesthetics and lessen psychological effects on the patient and family [31]. The impacted teeth can be temporarily retained as there may be a tendency for continued development and eruption during follow-up [37, 38, 42, 43]. Besides, it can prevent excessive alveolar ridge resorption after premature tooth loss and subsequent repair difficulty. Thus, we opted for further treatment of the impacted teeth until the boy becomes an adult whilst continuing periodical reviews of his condition. Studies have shown that tooth eruption is mainly related to the dental embryo, dental capsule, parathyroid-related protein (PT HrP), and various growth factors and cytokines [44]. If the associated embedded teeth erupt during the wearing of the removable denture or if discomfort is caused due to jaw development, the denture should be ground or remade [37].

RO patients can undergo permanent prosthetic restoration until adulthood. For the impacted teeth, extraction is recommended unless the patient chooses a removable denture restoration. OR patients usually suffer from severe bone resorption in the affected area; thus, the effects of removable dentures might not be good. Implants can improve the retention and stability of the prosthesis. Currently, implant-based prostheses can be used for rehabilitating all types of edentulous spans with predictable outcomes [45]. Although the alveolar bone quality of the affected area in patients with RO may show no significant abnormalities [46], it is often insufficient for implant restorations [47]. The problem should be evaluated carefully and can be resolved through bone grafting. Autotransplantation of premolars to the affected area through orthodontic treatment has also been reported [41], and the treatment results showed that good functional occlusion could be established and complicated restorative treatment could be avoided [13, 48, 49]. Ziegler proposed that the length of the root should reach two-thirds or three-quarters of the final length when auto-transplanted [48].

RO patients are often associated with gingival redness and swelling, caries, maxillofacial infections, irregular dentition, dentition defects, *etc.* [3], therefore, requiring multidisciplinary treatment for periodontics, endodontics, oral and maxillofacial surgery, pediatric stomatology, orthodontics, prosthodontics, and implants [24]. Further, psychological interventions might be required for patients with psychological effects due to premature tooth loss and abnormal tooth color and shape [21].

4. Conclusion

RO is a relatively rare non-genetic disease characterized by abnormalities in the development and eruption of teeth in a particular region within the mouth and can be diagnosed by clinical, radiographic and histopathological examinations. Early diagnosis is critical for timely intervention, selection of treatment strategies, follow-up, and monitoring [50, 51]. Early treatment can help prevent systemic diseases and reduce the complexity of future conditions. The treatment plan should be based on the severity of the disease as well as the patient's characteristics, such as age and functional and esthetic needs (Fig. 8). Treatment aims to preserve the patient's mastication, promote normal growth and development and reduce psychological impact [5], which can be achieved by multidisciplinary treatment, prolonged management and follow-up [37, 38], and compliance from the patients and their family members.

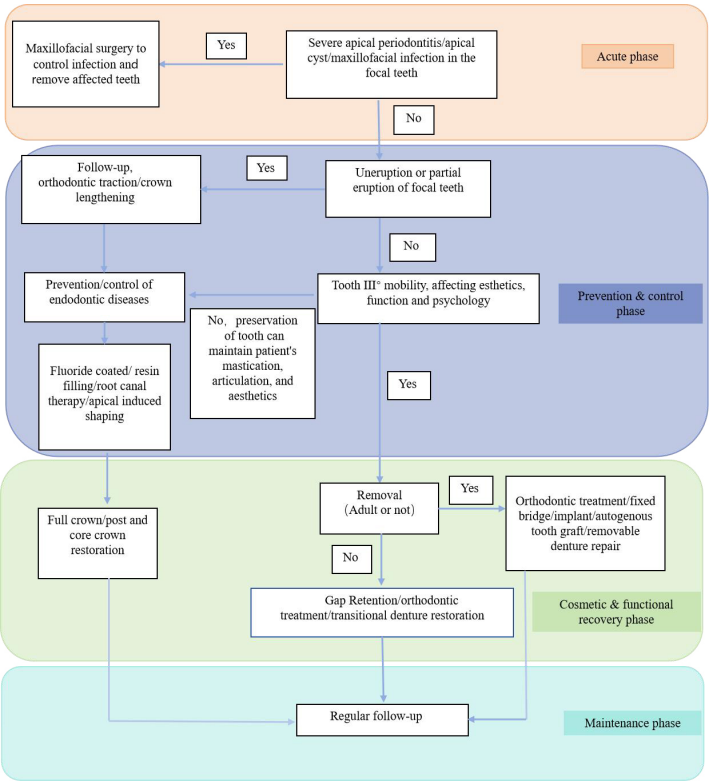

FIGURE 8. Clinical treatment scenarios for RO.

AVAILABILITY OF DATA AND MATERIALS

The data generated and/or analyzed during the current study are included in this published article.

AUTHOR CONTRIBUTIONS

LL—designed the research study. SBOY—performed the research. DMY, PZ—referred to and analyzed relevant literature review. DMY—summarized and wrote the manuscript. All authors read and approved the final manuscript.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

This study was approved by the Medical Ethics Committee of the Stomatology Hospital Affiliated to Nanchang University (AF/SG-04/1.0), with approval number 2018030. Informed consent was obtained by phone from the patient's parents before writing this retrospective case report.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

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