Case report

REGIONAL ODONTODYSPLASIA IN A NIGERIAN CHILD: A Case Report and Literature review

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ABSTRACT

Background: Regional odontodysplasia also known as 'ghost teeth' is a rare developmental disorder whose aetiology is unknown. The suspected factors linked with the condition are trauma, infections etc. It affects both primary and permanent dentition with a female predilection, usually localized to one segment of the jaw and involved teeth appear yellowish-brown, with rough surfaces and are prone to fracture. Radiographically, enamel and dentine are thinner than normal and lack demarcation, pulp chambers are large with short roots. Managements of these cases should be based on esthetics and functional needs as well as the degree of involvement of the tooth.

Objective: There is dearth of studies on this topic in Nigeria, hence we have decided to report this incidental finding of regional odontodysplasia seen in our center.

Case report: A 6-year-old girl presented to the Paediatric clinic with complaint of pain from a tooth in the upper right quadrant. On examination, presence of mobile 55 that appeared yellowish with rough surfaces without any carious lesion. Radiograph showed lack of distinction between enamel and dentine, large pulp chamber with short roots of tooth 55 and unerupted 16. Management was extraction of affected tooth with of stainless-steel crown placement on 16 after eruption. Patient is still being monitored.

Conclusion: Regional odontodysplasia although rare can present at any time. Management of regional odontodysplasia should be approached individualistically based on patient's presenting complaint.

Keywords: Developmental disorder, jaws, Regional Odontoplasia

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INTRODUCTION

Regional odontodyspalsia (RO) is a rare non-hereditary developmental anomaly of the tooth apparatus that affects the enamel, dentin, and cementum.[1] Although the aetiology of this dental anomaly is unknown, several factors have been postulated, such as local trauma, infection, teratogenic medications, irradiation, Rhesus incompatibility, hyperpyrexia, metabolic and nutritional imbalance. In both the primary and permanent dentition, this commonly affects

unilateral contiguous teeth. RO is thought to be slightly more common in females and the maxilla.[2,3] It is typically unilateral, however, it can also be bilateral and crosses the midline on rare occasions.[3]

The diagnosis of RO depends on clinical and radiographic findings and is assisted by histopathologic features. Clinically, the tooth affected with RO has an aberrant shape and an irregular mottled facial contour. The teeth appear to be hypoplastic or hypocalcified and are yellow or yellowish-brown in colour. Teeth may not erupt at all or may erupt later than expected. Even in the absence of dental caries, periapical infections and abscess formation are the most common clinical manifestations after the eruption of teeth affected with RO. The diseased teeth have aberrant morphologies and hypoplastic crowns on radiographs, and the enamel and dentine appear very thin, giving them a ghost-like appearance. Other pathognomonic radiological features include enlarged pulp chambers, short roots, open apices, and shell-like crowns.[4,5] On histology, there is reduction in the thickness of the dentin layer, expanded areas of interglobular dentin and extended pre-dentin is observed. In severe cases, the dentinal tubules are reduced in number, whereas in the milder cases the mantle dentin appears normal. The pulp often contains denticles and amorphous calcified materials.[6]
This article reports a case of an incidental finding of regional odontodysplasia affecting the right maxillary quadrant of a 6-year-old girl who presented to the University of Benin Teaching Hospital (UBTH).

CASE REPORT:

A 6-year-old girl presented to the Paediatric Dentistry clinic of the University of Benin Teaching Hospital, Benin City, Nigeria with a primary complaint of pain and mobility on tooth 55. The pain was sharp, spontaneous, prevented her from chewing on that side but did not disturb sleep. There was no previous history of dental visit. Her mother had no history of the disease, the child is the first of two children, and there was no history of genetic or dental disorders or anomalies in the family. The child had normal skin, hair and was in good general health, with no history of hereditary or acquired medical conditions. Intraoral examination revealed an abnormally shaped mottled tender tooth 55 with grade III mobility and the presence of a sinus (Fig. 1).

Figure 1: A clinical photograph showing a discolored 55 with mottled appearance different from the other teeth

Periapical radiographs revealed tooth 55 and unerupted 16 on the maxillary right quadrant had no distinction between the enamel and dentine with wide pulp chambers and short roots giving the “ghost-like appearance” (Fig. 2).

Figure 2: A digital periapical radiograph showing 55 and unerupted 16 showing mottled appearance, no distinction between enamel and dentine, large pulp chambers and short roots.

There was a delay in the eruption of 16 as 26, 36 and 46 had all fully erupted on the arch. With the radiographic finding seen, a panoramic view was taken to assess if other quadrants were affected and the integrity of the developing permanent tooth buds. This confirmed the abnormality affecting only the 55 and 16 teeth (Fig. 3).

Figure 3. Orthopantomogram showing the presence of ghost teeth on the right maxillary quadrant. Also note the presence of 26, 36, and 46 on the arch.

Based on clinical and radiographic findings, regional odontodysplasia was diagnosed. With parental consent, tooth 55 was extracted under local anaesthesia and planned for a space maintainer. Mother was advised on the condition of the unerupted 16 and a plan of placing a stainless-steel crown as soon as it erupts was emphasized. The patient and mother were educated on the importance of regular recall visits for assessments, dietary counselling, and oral hygiene instructions were given and the patient was motivated.

DISCUSSION

Regional odontodysplasia is a rare developmental anomaly with an unknown cause, with just about 150 cases reported in the English literature Amelogenesis and dentinogenesis imperfecta are frequently mistaken as regional odontodysplasia and unlike the latter, all the former affects the entire dentition and do not show a segmental distribution.[3] It can be found in both the primary and permanent dentition, with the maxilla showing a stronger predilection.[7] The condition has a female to male ratio of 1.4:1.[8] This report is similar to many other studies as odontodysplasia is seen to affect the primary and permanent dentition in the maxillary quadrant in a young female.[2,9] Although some reports suggest that the left side of the maxilla is commonly affected, the right posterior maxillary quadrant was affected in this case.[3,6,9]

Several factors have been proposed as contributing to the aetiology of RO,[8,10] There was no known contributing factor for regional odontodysplasia in this case, and the patient did not have any known systemic condition linked with regional odontodysplasia. In previous studies, the most common clinical features were yellowish-brown colour and delayed eruption of permanent teeth, with radiographs showing ghost-teeth and poorly developed buds.[9–11] However, in this case, the features seen were yellowish mottled teeth, mobility and pain from the affected teeth, delayed eruption of a permanent first molar on the affected side, and ghost-teeth characteristics seen on the radiograph.

Although this was an accidental finding because the patient's presenting complaint was pain, clinical and radiological methods were used to make the diagnosis of regional odontodysplasia. Treatment for regional odontodysplasia varies depending on the severity of the disorder and can include conservative restorations, root canal treatment, and extraction, with the extraction of the affected teeth followed by prosthetic rehabilitation being the most common treatment because the teeth tend to develop pathosis even after conservative treatment.[9] The treatment of choice, in this case, was scaling and polishing with fluoride therapy as part of general management and extraction of the affected primary tooth, with a plan for space management and close monitoring of tooth 16 until it erupts into the oral cavity, based on clinical presentation. Another study, on the other hand, used conservative non-surgical periodontal and fluoride therapy with a follow-up that showed marked improvement this may be due to

the fact that the presenting complaint was gingival swelling with delayed eruption of teeth.[12] In addition to the treatment done for this case, the importance of regular follow-up was emphasized, and the mother was thoroughly educated about the condition.

CONCLUSION

The purpose of presenting this case is to raise awareness about the likelihood of encountering this rare disease in our environment. Also, the fact that therapeutic considerations should be based on the degree of the abnormality and condition of affected teeth at the time of presentation as well as the functional and esthetic needs of each case.

Recommendation

We recommend that full mouth radiographs in the form of an orthopantomogram (OPG) be taken at the patient's first visit as this can help identify any problems with dental hard tissue structures and also serve as a focal point for early diagnosis of regional odontodysplasia as well as prompt treatment.

Conflict of interest: No conflict of interest

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