

Case Report

Corresponding author

Makoto Matsubara

Department of Oral and Maxillofacial Surgery

Division of Oral Pathogenesis and Disease Control

Asahi University School of Dentistry

Hozumi 1851, Mizuho

Gifu 501-0296, Japan

Tel. +81 58 329 1472

Fax: +81 58 329 1472

E-mail: m-matsu@dent.asahi-u.ac.jp

Volume 1 : Issue 1

Article Ref. #: 1000DOJ1101

Article History

Received: September 15th, 2014

Accepted: September 26th, 2014

Published: September 27th, 2014

Citation

Matsubara Y, Matsubara M, Iinuma M, Tamura Y. Clinical management of regional odontodysplasia. *Dent Open J.* 2014; 1(1): 1-4. doi: [10.17140/DOJ-1-101](https://doi.org/10.17140/DOJ-1-101)

Copyright

©2014 Matsubara M. This is an open access article distributed under the Creative Commons Attribution 4.0 International License (CC BY 4.0), which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.

Clinical Management of Regional Odontodysplasia

Yuiko Matsubara¹, Makoto Matsubara^{2*}, Mitsuo Iinuma¹ and Yasuo Tamura¹

¹Department of Pediatric Dentistry, Division of Oral Structure, Function and Development, Asahi University School of Dentistry, 1851 Hozumi, Mizuho, Gifu 501-0296, Japan (Director: Prof. Yasuo Tamura)

²Department of Oral and Maxillofacial Surgery, Division of Oral Pathogenesis and Disease Control, Asahi University School of Dentistry, Hozumi 1851, Mizuho, Gifu 501-0296, Japan (Director: Prof. Michio Shikimori)

ABSTRACT

Here we present a case of a 7-year-old Japanese girl with regional odontodysplasia affecting tooth numbers 24, 25, 26, and 27. No other teeth were affected. Partially erupted tooth number 26 had a rough enamel surface and was yellow-brown in color. Tooth numbers 24, 25, and 27 had not erupted. Radiographically, numbers 24, 25, 26, and 27 seemed to be undeveloped with large pulp chambers, short roots, and open apices, and exhibited the so-called ghost-like appearance. Histopathologically, number 26 had uneven thin enamel and dentin, revealing hypoplasia and hypocalcification, and irregular dentinal tubes. The patient was followed up for 6 years and had similarity normal occlusion.

KEYWORDS: Odontodysplasia; Ghost teeth; Hypoplastic permanent teeth.

INTRODUCTION

Odontodysplasia is a rare developmental anomaly in tooth formation generally observed in young patients. This disease is characterized by the malformation and hypocalcification of both the enamel and dentin, asymmetrically and locally. Since the initial description by McCall and Wald,¹ approximately 125 cases have been reported in the dental literature worldwide. The disease is variously described as odontodysplasia,² localized arrested tooth development,^{3,4} odontogenesis imperfecta,⁵ and unilateral dental malformation.⁶ Rushton⁷ termed the disease “ghost teeth” due to the ghost-like appearance of the teeth radiographically. Here we report a case of regional odontodysplasia in a 7-year-old Japanese girl. The patient was followed up for 6 years and had normal occlusion.

CASE REPORT

This 7-year-old girl was referred to Asahi University Hospital in Gifu, Japan, for diagnosis and treatment of an unusual enlargement in the right maxilla. Her chief complaint was pain and swelling in the upper right molar region.

According to the patient’s mother, tooth numbers 64 and 65 erupted at 6 months after birth and were very different from those on the other side of the maxilla, with altered morphology and a yellowish color. They were quickly destroyed by caries, which was often accompanied by abscess formation.

At 6-years of age, numbers 64 and 65 were extracted due to pain and swelling at a private dental clinic. Two weeks after the extraction of 64 and 65, tooth number 26 began to erupt. The external appearance of 26 was similar to that of 64 and 65, and associated with some swelling and redness. No undisputed cases of regional odontodysplasia were observed in other family members. Her general condition was satisfactory. Comprehensive extraoral and head-and-neck examinations were non-contributory, except for a slight depression of the right cheek (Figure 1).



Figure 1: Facial and neck photograph taken at 7 years and 7 months

When she first visited this hospital, tooth number 26 was still present, and characterized by a rough enamel surface and yellowish coloration with high mobility. Numbers 24 and 25 had not erupted (Figure 2). Radiographically, 24, 25, 26, and 27 exhibited an altered morphology associated with little demarcation between the enamel and dentin, and were undeveloped with large pulp chambers, short roots, and open apices, and appeared ghost-like radiographically. Tooth numbers 24, 25, and 27 were still inside the bone. The number and morphology of the remaining dentition were normal (Figure 3).

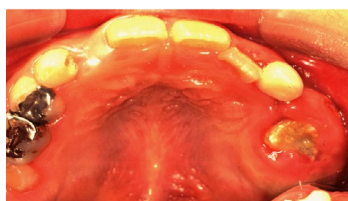


Figure 2: Intraoral photograph of maxilla taken at 7 years and 7 months



Figure 3: Orthopantomograph taken at 7 years and 7 months

We judged that 26 could not be preserved, and it was therefore extracted under local anesthesia (Figure 4). The remaining dentition was quite normal. After extraction, the patient was fitted with a retainer to maintain the space of 26 (Figure 5).

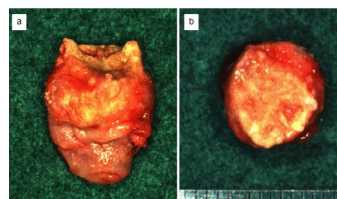


Figure 4: Extracted tooth a) lateral surface b) occlusal surface



Figure 5: Intraoral findings 1 month after tooth extraction

Histopathologic examination of extracted tooth number 26 revealed uneven thin enamel and dentin, indicating hypoplasia and hypocalcification, irregular dentinal tubes, and marked reduction of the dentinal tube number in the crown portion (Figure 6).

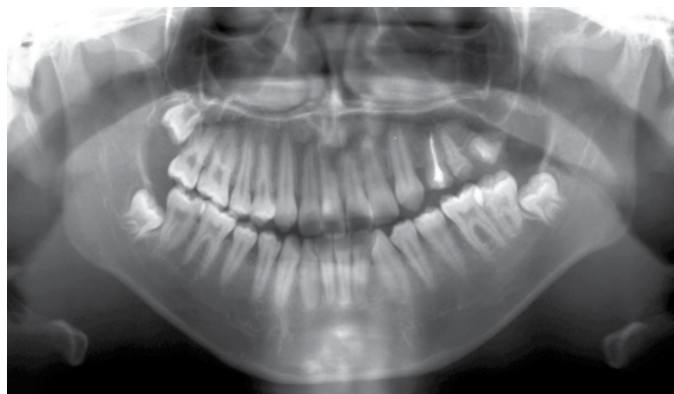


Figure 6: Orthopantomograph taken at 13 years.

DISCUSSION

According to the literature,⁷ regional odontogenesis is slightly more prevalent in females⁸ and slightly more often involves the maxillary arch. In this case, the patient was female and the maxillary arch was affected. Both the dentition (primary and permanent dentition) were affected, and the age of diagnosis was during primary tooth eruption as well as during mixed dentition.⁹

Local infection, trauma, oral ischemia, somatic mutation, irradiation, metabolic or vitamin deficiency have been discussed as possible etiologies of this developmental disturbance.^{6,10,11-15}

Possible causes of the present case could not be determined. Rushton⁸ considered that regional odontodysplasia could be caused by either a single act of damage with resulting subsequent abnormalities or continuing insult. It has been argued, however, that a single act of damage would have to occur in utero and it is difficult to envisage such damage extending from the deciduous to the permanent dentition, with the occasional tooth in a quadrant remaining unaffected.¹⁶ Activation of a latent virus in the odontogenic epithelium has also been suggested as a cause for the disorder,^{7,17,18} but no virus particles have been identified.¹³

Local infection has also been cited,¹² but the casual relationship between infection and hypoplasia is debatable.¹³

Similarly, local trauma has been implicated.^{14,18} If trauma was definitely related to a specific tooth during development, however, then such a case probably should not be classified as regional odontodysplasia.¹⁵

Metabolic or vitamin deficiencies have also been advanced as causative factors, but it is unlikely that only specific teeth would be affected and not others at similar stages of development.¹⁴ Irradiation can cause damage to developing teeth,^{7,14} but in such cases a history would be available, and all the developing teeth in the treated area would be affected. The involvement of selected teeth in regional dysplasia leaving adjacent teeth unaffected makes radiation an unlikely cause.

The somatic mutation theory,^{7,17} does not account for cases in which unaffected teeth have developed from the distal extension of a mesially affected dental lamina, and in addition, when teeth in both jaws are affected the chances of multiple mutations in the same patient are highly unlikely.¹⁹

A hereditary factor can be discounted because of the lack of evidence of such a history in cases reported to date. The possibility of maternal illness, drug therapy, or both as etiologic factors was suggested by cases reported in children whose mothers had pneumonia¹⁴ or toxemia¹⁶ during pregnancy or who took diuretics,²⁰ tranquilizers, or hormones during the first trimester.⁹

Although a relationship between drugs and regional odontodysplasia has not been established, increased awareness of the possible effects of such harmful agents is required. It is well established that a great variety of drugs taken during pregnancy can cause congenital malformations, such as warfarin taken during the first trimester,²¹ ethanol causing fetal alcohol syndrome,²² cocaine producing congenital malformations,²² aromatic retinoids used for severe skin disease,²³ and angiotensin-converting enzyme inhibitors for hypertension.²⁴ Children of woman with epilepsy also appear to have increased evidence of congenital malformations; it is not certain whether this is due to drug therapy, but it seems likely.²²

This disease can be distinguished from other disturbances of odontogenesis because all of the histologic elements of the dental organ are abnormal in the affected teeth, while others in the same individual are normal.

The treatment plan for this patient was removal of tooth numbers 24, 25, and 27 after eruption if the hypoplasia and hypocalcification could not be remedied. In many cases of odontodysplasia, the teeth cannot be preserved.²⁵

The histopathologic features and clinical and radiographic aspects were analyzed and the diagnosis of regional odontodysplasia was established.

CONCLUSION

We reported a case of regional odontodysplasia in a 7-year-old Japanese girl. The patient was followed up for 6 years and had similarity normal occlusion.

CONSENT

The patient has provided written permission for publication of the case details.

CONFLICTS OF INTEREST

We have no conflicts of interest to declare.

REFERENCES

1. McCall JO, Wald SS. Clinical dental roentgenology. 2nd ed, Philadelphia and London, US and UK: *W. B. Saunders Co.*; 1947: 150.
2. Zilo JA, Ringler D, Mandel L. Single tooth odontodysplasia. Case report. *NY State Dent J.* 2013; 79: 35-37.
3. Suher T, Jump EB, Landis RL. Localized arrested tooth development. *Oral Surg Oral Med Oral Pathol.* 1953; 6: 1305-1314. doi: [http://dx.doi.org/10.1016/0030-4220\(53\)90262-X](http://dx.doi.org/10.1016/0030-4220(53)90262-X)
4. Bang E, Kjaer I, Christensen LR. Etiologic aspects and orthodontic treatment of unilateral localized arrestedtooth-development combined with hearing loss. *Am J Orthod Dentofacial Orthop.* 1995; 108: 154-161. doi: [http://dx.doi.org/10.1016/S0889-5406\(95\)70078-1](http://dx.doi.org/10.1016/S0889-5406(95)70078-1)
5. Edward A, Neupert III, John MW. Regional odontodysplasia presenting as a soft tissue swelling. *Oral Surg Oral Med Oral Pathol.* 1989; 67: 193-196. doi: [http://dx.doi.org/10.1016/0030-4220\(89\)90329-0](http://dx.doi.org/10.1016/0030-4220(89)90329-0)
6. Bergman G, Lysell L, Pindborg JJ. Unilateral dental malformation. Report of two cases. *Oral Surg Oral Med Oral Pathol.* 1963; 16: 48-60. doi: [10.1016/0030-4220\(63\)90363-3](http://dx.doi.org/10.1016/0030-4220(63)90363-3)
7. Rushton MA. Odontodysplasia."Ghost Teeth". *Brit Dent J.* 1965; 119: 109-113.
8. Lustmann J, Klein H, Ulmanky M. Odontodysplasia. Report of two cases and review of the literature. *Oral Surg.* 1975; 39: 781-793.
9. Cahuana A, Gonzalez Y, Palma C. Clinical management of regional odontodysplasia. *Pediatr Dent.* 2005; 27: 34-39.
10. Gomes MC, Modesto A, Cardoso AS, Hespanhol W.

Regional odontodysplasia: report of a case involving two separate affected areas. *J Dent Child*. 1999; 66: 203-207.

11. Ozer L, Cetiner S, Ersoy E. Regional odontodysplasia: report of a case. *J Clin Pediatr Dent*. 2004; 29: 45-48.

12. Hintz CS, Peters RA. Odontodysplasia. Report of an unusual case and a review of the literature. *Oral Surg Oral Med Oral Pathol*. 1972; 34: 744-750. doi: [10.1016/0030-4220\(72\)90292-7](https://doi.org/10.1016/0030-4220(72)90292-7)

13. Gardner DG, Sapp JP. Regional odontodysplasia. *Oral Surg*. 1973; 35: 351-365.

14. Zegarelli EV, Kutscher AH, Applebaum E, Archard HO. Odontodysplasia. *Oral Surg Oral Med Oral Pathol*. 1963; 16: 187-193.

15. Abrams AM, Groper J. Odontodysplasia, Report of three cases. *J Dent Child*. 1966; 33: 353-362.

16. Ferguson JW, Geary CP. Regional odontodysplasia. *Aust Dent J*. 1980; 25: 148-151. doi: [10.1111/j.1834-7819.1980.tb03705.x](https://doi.org/10.1111/j.1834-7819.1980.tb03705.x)

17. Hermam NG, Moss SJ. Odontodysplasia: report of case. *J Dent Child*. 1977; 44: 52-54.

18. Bergman G, Lysell L, Pindborg JJ. Unilateral dental malformation. Report of two cases. *Oral Surg Oral Med Oral Pathol*. 1963; 16: 48-60.

19. Walton JK, Wirkop CJ Jr., Walker PO. Odontodysplasia. Report of three cases with vascular nevi overlying the adjacent skin of the face. *Oral Surg Oral Med Oral Pathol*. 1978; 46: 676-684.

20. Alexander WN, Lilly GE, Irby WB. Odontodysplasia. *Oral Surg Oral Med Oral Pathol*. 1966; 22: 814-820.

21. Warkany J. Warfarin embryopathy. *Teratology*. 1976; 14: 205-209. doi: [10.1002/tera.1420140211](https://doi.org/10.1002/tera.1420140211)

22. Farmer RE. Teratogenic drugs: an update. *Adverse Drugs Reaction Bulletin*. 1993; 161: 607-610.

23. Dai WS, LaBraico JM, Stem RS. Epidemiology of isotretinoin exposure during pregnancy. *J Am Acad Dermatol*. 1992; 26: 595-606. doi: [10.1016/0190-9622\(92\)70088-W](https://doi.org/10.1016/0190-9622(92)70088-W)

24. Barr MJr., Cohen MMJr. ACE inhibitor fetopathy and hypocalvaria: the kidney-skull connection. *Teratology*. 1991; 44: 485-495. doi: [10.1002/tera.1420440503](https://doi.org/10.1002/tera.1420440503)

25. van der Wal JE, Rittersma J, Baart JA, van Waal I. Regional odontodysplasia report of three cases. *Oral Maxillofac Surg*. 1993; 22: 356-358. doi: [http://dx.doi.org/10.1016/S0901-5027\(05\)80667-8](https://doi.org/http://dx.doi.org/10.1016/S0901-5027(05)80667-8)