Idiopathic Oligodontia in Primary Dentition: Case Report and Review of Literature

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Idiopathic oligodontia of the primary dentition is very rare. This is a case report of a 2 year and 6 month old boy, who presented with fourteen missing primary teeth. Review of previous case reports is done and the management of these cases is discussed.

Key words: *Oligodontia, primary teeth, Idiopathic.*
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INTRODUCTION

According to the widely accepted definition as given by Stewart,[1] oligodontia means agenesis of numerous teeth (more than 6) whereas hypodontia is defined as, absence of one or few teeth and anodontia, complete absence of teeth. Oligodontia of permanent dentition is common and is considered as a variant of normal pattern.[2]

Hypodontia of primary dentition is rare and the reported prevalence depends on various factors like geographic location (0.1% to 0.9% bases on studies in Iceland, Scandinavia and Britain) and the study methods used.[3,4,5] A higher incidence of 5% has been reported in Japanese population.[6] The most common missing teeth are maxillary lateral incisors and mandibular central and lateral incisors.[7] The causes of oligodontia are hereditary, trauma, infection, radiation, metabolic disorders and idiopathic. It can occur alone or in association with syndrome like ectodermal dysplasia.[8] Oligodontia of primary dentition can cause impaired growth of alveolar process, reduced lower facial height, pseudoprognathism, speech impairment and deep bite, all of which can have a physiological and psychosocial impact on the individual.[9] This is a case report of fourteen missing primary teeth with discussion on the management.

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CASE REPORT

A 2 year and 6 month old boy was referred to Department of Pediatric Dentistry, Christian Dental College, Ludhiana with a chief complaint of several missing teeth. The child was in good health and the medical history did not reveal any systemic disease. Child was born to non-consanguinous parents. Mother reported an uneventful pregnancy. The patient's two elder siblings girls aged 9 and 7 years did not have any missing teeth. The family history was not significant. Mother reported no prior history of trauma, extraction or exfoliation of teeth in this child.

On examination there was no abnormality of skin, nails and hair. There was no history of sweating abnormality. His systemic examination was normal. An intra-oral examination revealed the presence of maxillary right and left central incisors, right and left primary second molars and mandibular right and left primary second molars. The teeth were of normal size, shape and color. Dental caries was present in the cervical region of the upper right and left central incisors (Figure 1).

The panoramic radiograph revealed presence of four anterior permanent tooth buds in the maxillary arch and the

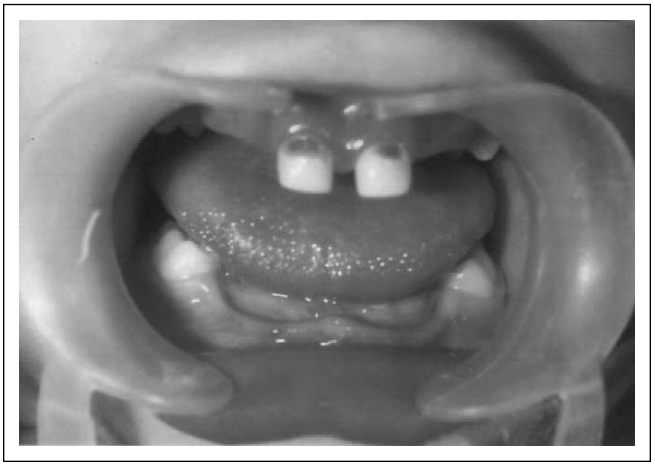

Figure 1. An intra-oral examination revealed the presence of maxillary right and left central incisors, right and left primary second molars and mandibular right and left primary second molars.

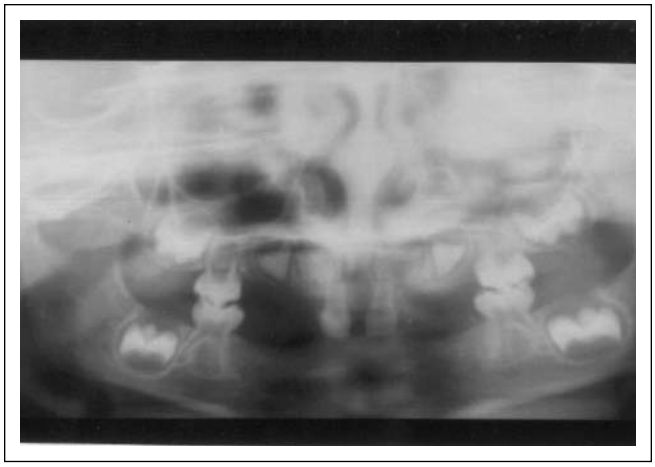

Figure 2. The panoramic radiograph revealed presence of four anterior permanent tooth buds in the maxillary arch and the corresponding expected tooth buds in the mandibular arch were absent.

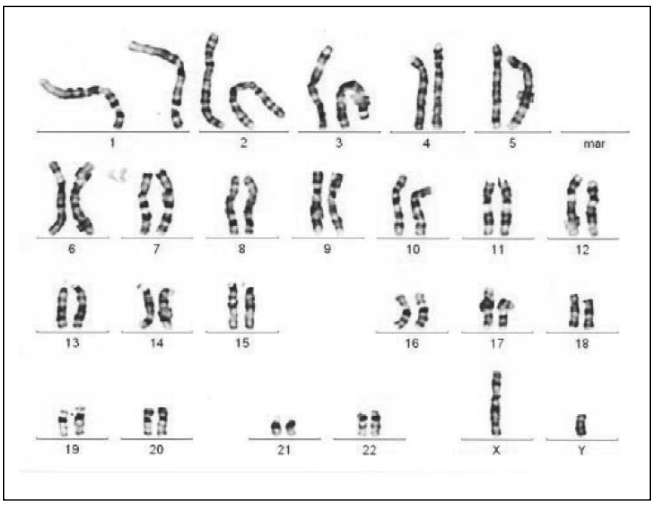

Figure 3. Normal male karyotype (46, XY).

corresponding expected tooth buds in the mandibular arch were absent. It also revealed the presence of all the four permanent molar tooth buds. The alveolar ridge was very thin. (Figure 2) Karyotyping analysis done to rule out known structural chromosomal abnormalities, which showed a normal male type (46,XY) (Figure 3). The treatment included restoration of the caries lesion, prosthesis in the form of simple acrylic partial dentures and dental health education.

DISCUSSION

This case report is unique in the following aspect: it reports maximum number of missing primary teeth, missing primary first molars, missing permanent tooth buds and absence of clinical features of syndromes associated with oligodontia of primary dentition. Daugaard-Jensen *et al*[10] have reported one case with 14 missing teeth in their study of 193 cases. But there is no mention as to whether this particular case is syndrome associated or not. Ringqvist and Thilander[11] have reported a case with seventeen missing teeth in their study in 5513 children. But the age of the child and type of dentition is not clear from the data given. Recently there has been a reported case of idiopathic oligodontia in primary dentition by Shashikiran *et al*[12] with nine missing teeth.

Idiopathic agenesis of primary molars is extremely rare.[13,14] In this patient all the first primary molars were absent, which has not been reported earlier. The radiographic evidence also did not show any embedded primary molar tooth buds. It also revealed presence of upper permanent anterior tooth buds and absence of corresponding expected lower tooth buds, which is rare. By the age of three years, mandibular central and lateral incisors, canines and first molars and maxillary central and lateral incisors, canines and first molars permanent tooth buds should be present radiographically.[15] In this case only upper anterior permanent tooth buds were present. The lower tooth buds may be congenitally missing or may appear at a later stage.

There was absence of clinical features of syndromes known to be associated with oligodontia: the patient did not show any features of syndromes like Reiger's syndrome (defects of iris, cornea, anterior chamber of eye and myotonic dystrophy), Marshall's syndrome (defects of eye, deafness, facial characteristics like depressed nasal bridge, frontal bossing and hypertelorism), Lacrimo-auriculo-dento-digital syndrome (abnormalities of lacrimal gland, naso-lacrimal duct, ears and digits), Down's syndrome (mental retardation, abnormalities of eyes) and ectodermal dysplasias. The karyotyping done in this case showed a normal male type (46 XY). Therefore, this is a case of idiopathic Oligodontia with maximum missing teeth in primary dentition reported so far. Ooshima *et al*[2] reported a case of oligodontia in primary dentition with all permanent tooth

Table 1. Clinical detail of cases with idiopathic oligodontia in primary dentition reported so far.

Author/year	Age/Sex	No. of Missing Teeth	Teeth Missing	
Ooshima T et al 1988	2yr/M	8	54 53 xx xx	xx 62 63 64
			84 83 xx xx	xx xx xx 74
Shashikiran et al 2002	3yr/M	9	54 xx xx 51	61 xx xx 64
			84 xx xx 81	xx xx xx 74
Present case report	2? yr/M	14	55 xx xx xx 51	61 xx xx xx 65
			85 xx xx xx xx	xx xx xx xx 75

buds present including the corresponding successors of the congenitally missing primary teeth.

Several factors like trauma, infection of the developing tooth bud, radiation overdose, systemic conditions like rickets and syphilis, several intrauterine disturbances have been proposed as etiological factors.[16] Ranta and Maroto have suggested a hereditary cause for oligodontia.[17,18] Nieminen *et al* have demonstrated deletion of *MSX1* gene (essential for normal oral and tooth development) on chromosome 4 in patients with Wolf-Hirschhorn Syndrome who have oligodontia.[19] Jorgenson suggested physical interruption of dental lamina as a causative factor of oligodontia as seen in oro-facio-digital syndrome.[20]

Several studies have shown strong correlation between the agenesis of primary teeth and agenesis of corresponding permanent teeth.[21,22] In the study by Grahnen and Granath 75% of the cases with hypodontia in primary dentition showed the same condition in the permanent dentition.[13] In this patient upper anterior permanent tooth buds are present. This case needs to be followed up to determine the correlation between the agenesis of primary and permanent dentition. If there is agenesis in the primary but not in the permanent dentition the etiological factor cannot be a defect in the down growth of the dental lamina. If on the other hand there is agenesis of both dentitions, the condition may be due to ectodermal mucosal defect. To conclude in this aspect our case needs a follow up, in terms of shape and structure of the developed teeth and other ectodermal manifestations.

Interdisciplinary management is essential in these cases for optimal outcome. It is important to protect the teeth present in these very young patients. Preventive restorative procedures can be done before undertaking further prosthesis. Any existing caries lesion should be restored. Further management depends on the age of the patient. Between 0-6 years removable partial denture is recommended, keeping in view the growing age of the patient. Rigid fixed prosthesis is contraindicated at this age. In children between 7-12 years prosthesis and restorations provided during the earlier treatment should be maintained and enhanced. Rigid or fixed prosthesis should be used with caution and monitored. Minor tooth movement and selective extraction of teeth should be considered for accommodating prosthesis.

In adolescent age group prosthetic replacement of missing teeth is appropriate. Conservative rigid or fixed prosthesis may be used if growth is completed. Comprehensive orthodontic tooth movement and selective extraction of teeth can be done. Implant is recommended only for the missing lower anterior teeth. The patient will be monitored every six months to determine the need to refit or remake his complete denture. This will be a transitional period during which the patient response to his prosthesis will be evaluated. Further follow up for evaluating the role of multiple missing primary teeth in the development of dento-facial structures needs to be evaluated. Based on this, a long-term treatment protocol for idiopathic Oligodontia in primary dentition and its corresponding permanent successors needs to be undertaken. The treatment guidelines suggested in this paper are an attempt to discuss the options available to the pediatric dentist for the oral rehabilitation of children with oligodontia.

CONCLUSION

This case highlights the role of a pediatric dentist in the thorough clinical evaluation, early and correct diagnosis, optimal management along with an interdisciplinary team depending on the age of patient and careful monitoring during the follow up in patients with Oligodontia in both primary and permanent dentition.

REFERENCES

1. Stewart RE, Barber TK, Troutman KC et al. The dentition and anomalies of tooth size, form, structure and eruption. In Pediatric Dentistry. Ed. St. Louis, The CV Mosby 91, 1982.
2. Ooshima T, Sugiyama K, Sobue S. Oligodontia in the primary dentition with permanent successors: report of case. J Dent Child 55: 75–77, 1988.
3. Moller P. Oral health survey of preschool children in Iceland. Acta Odontol Scand 21: 47–97, 1963.
4. Ravn JJ. Aplasia, supernumerary teeth and fused teeth in the primary dentition. An epidemiological study. Scand J Dent Res 79: 1–6, 1971.
5. Brook AH. Dentine anomalies of number, form and size: their prevalence in British School children. J Int Assoc Dent Child 5: 37–53, 1974.
6. Saito TA. Genetic study on degenerative anomalies of deciduous teeth. Jpn J Mum Genet 4: 27, 1959.
7. Braham RL, Morris ME. Defect of the teeth and tooth bearing structures. In Text Book of Pediatric Dentistry. 2nd Ed. CBS Publishers and Distributors, 2–109, 1990.
8. Schalk-vander weide Y, Beemer FA, Faber JAG, Bosman F. Symptomatology of patients with oligodontia. Oral Rehabil 21: 246–261, 1994.
9. Kloeppel JG. Die Grobenentwicklung des Oberkiefers bei Fallen von Zahnunterzahl. Fortschr Kieferorthop 18: 85–89, 1957.
10. Daugaard-Jensen J, Nodal M, Skovgaard LT, Kjaer I. Comparison of pattern of agenesis in the primary and permanent dentition in population characterized by agenesis in the primary dentition. Int J Ped Dent 7: 143–148, 1997.
11. Ringqvist M, Thilander B. Frequency of hypodontia in an orthodontic material. Sven Tandl Tidskr 62: 535–541, 1969.
12. Shashikiran ND, Karthik V, Subbareddy VV. Multiple congenitally missing primary teeth: report of a case. Pediatr Dent 24: 149–152, 2002.
13. Granhnen H, Granath LE. Numerical variations in primary dentition and their correlation with the permanent dentition. Odont Revy 12: 348–357, 1961.
14. Bennet CG, Ronk SL. Congenitally missing primary teeth: report of case. J Dent Child 47: 346–348, 1980.
15. Kupietzky A, Houpt M. Hypohydrotic ectodermal dysplasia: characteristics and treatment. Quintessence Int 26: 285–291, 1995.
16. Arya BS, Savara BS. Familial partial anodontia. Rev Stomatoodontol Nord Frq 27: 197, 1972.
17. Ranta R. Hereditary agenesis of ten maxillary posterior teeth: a family history. J Dent Child 52: 125–127, 1985.
18. Maroto MR, Azcorra GV, Arellanos RO, Martin MTB. Un caso de agenesia de incisivios permanentes y de sus correspondented piezas deciduas. Rev Act Estomatologica espanola 47: 39–42, 1987.
19. Nieminen P, Kotilainen J, Aalto Y, Knuutila S, Pirinen S, Thesleff I. MSX1 gene is deleted in Wolf-Hirschhorn Syndrome patients with oligodontia. J Dent Res 82: 1013–1017, 2003.
20. Jorgenson RJ. Clinician's view of Hypodontia. J Am Dent Assoc. 101: 283–86, 1980.
21. Whittington BR, Durward CS. Survey of anomalies in primary teeth and their correlation with permanent dentition. New Zealand Dent J 92: 4–8, 1996.
22. Davis PJ, Darvell BW. Congenitally missing permanent mandibular incisors and their association with missing primary teeth in the southern Chinese (Hongkong). Comm Dent Oral Epidemiol 21: 162–164, 1993.