

Case Report

NONSYNDROMIC OLIGODONTIA WITH ANKYLOGLOSSIA: A RARE CASE REPORT

Arihant Jain¹, Priyanka Thakur¹, Sahil Sarin²

Junior Resident, ¹Department of Pedodontics and Preventive Dentistry, ²Prosthodontics, HP Government Dental College, Shimla, Himachal Pradesh

Abstract:

Agenesis of teeth is one of the most common human developmental anomalies. Oligodontia is a condition in which there is absence of six or more teeth, which itself is a rare condition. Oligodontia is associated with many syndromes. There is no much literature on oligodontia with ankyloglossia. This is case report of a 9 year old girl child, who had missing permanent mandibular anterior teeth and these was also presentation of ankyloglossia. The case report discusses the treatment options and management of the case at this age considering the growth left in the patient and psychologic effect that it might have on the patient.

Keywords: Oligodontia, Ankyloglossia, Nonsyndromic.

Corresponding Author: Dr. Arihant Jain, Department of Pedodontics and Preventive Dentistry, Email: dr_arihant_87@yahoo.com

This article may be cited as: Jain A, Thakur P, Sarin S. Nonsyndromic Oligodontia with Ankyloglossia: A Rare Case Report. J Adv Med Dent Scie Res 2015;3(1):143-147.

INTRODUCTION

Agenesis of teeth is one of the most common human developmental anomalies.¹⁻³ Hypodontia is the congenital agenesis of 5 or fewer permanent teeth, excluding third molars; oligodontia (also called severe hypodontia)^{1,4} is the congenital agenesis of 6 or more permanent teeth, excluding third molars;^{1,5} and anodontia is the congenital agenesis of all deciduous and/or permanent teeth.⁶

The dental agenesis is the result of disturbances in the stages of initiation and proliferation during the formation of teeth. Its etiology is associated with environmental factors such as infections, trauma, chemotherapy, radiotherapy and genetic causes.^{1,7}

The incidence of missing permanent teeth (excluding third molars) has been reported to vary from 2.6 to 11.3% depending on

demographic and geographic profiles, whereas the incidence of missing primary teeth is considerably low.⁷ Oligodontia is relatively a rare condition, probably affecting about 0.1 to 0.2% of the population.⁸ It may occur as a part of a syndrome or as an isolated condition that has been linked to mutations of the MSX1 and PAX9.⁹ It affects females more often than males, with a gender ratio of 3:2.¹⁰

The most commonly used method of diagnosis of dental anomalies is clinical examination accompanied by radiographic examination. The periapical and the panoramic radiographs are generally used for the radiographic diagnosis of dental agenesis.¹¹⁻¹³

There are several treatment options for adult and young patients with agenesis, although there are few studies demonstrating treatment in pediatric patients.¹⁴ A specialist with the

patient together must make the decision regarding treatment, since it is based not only on which teeth are missing, but also on the arch length, on the position of the incisors and lips, and on the aesthetic profile. The early diagnosis and treatment are important to improve masticatory function, speech, and self-appearance to reduce the psychosocial impact.^{14,15}

CASE REPORT:

A 9-year, 7-month-old Asian girl, accompanied by her parents, reported to the department of Pedodontics and Preventive dentistry at H.P. govt. dental college and hospital, Shimla (India) in may, 2014. Her chief complaint was missing lower front teeth. She was in healthy condition, with a normal physical body appearance, normal hair, eyebrows, fingers, nails, skin, and sweat glands, ruling out ectodermic dysplasia as the etiology. He had no history of trauma and tooth extractions, and his parents and siblings were dentally and medically normal. Further history revealed no teeth ever erupted in this region of the jaw, which suggests even the deciduous incisors were congenitally missing.

Extraoral examination revealed, she had a convex facial profile, obtuse nasolabial angle, marked labiomental fold with lip trap and reduced lower facial height (Figure 1 and 2). Facial symmetry was good and maintained on mandibular opening. The patient's maximal interocclusal space upon opening was 43-45 mm. No clicking or crepitus of the TMJs was observed. The muscles of mastication were not sensitive to palpation. The patient denied any facial or neck muscle pain.

Intraoral clinical examination revealed absence of mandibular incisors and canines (Figure 3) and presence of ankyloglossia which was classified as Class II by utilizing Kotlow's assessment and was able to protrude the tongue up to the lower lip but her speech was not affected. The patient was

in the mixed dentition stage and had a good ratio of transverse arches, Class II Division 1 malocclusion, and proclination of the permanent maxillary central incisors (Figure 4 and 5). Panoramic radiography indicated the absence of tooth germs of the permanent mandibular central incisors, lateral incisors, and the canines. The remaining permanent teeth were developing normally (Figure 6).



Figure 1: Front profile



Figure 2: Lateral profile



Figure 3: Occlusion: front view



Figure 4: Maxilla: occlusal view



Figure 5: Mandible: occlusal view



Figure 6: OPG

The following problems were identified: (1) missing permanent mandibular incisors and canines, (2) proclination of the permanent maxillary central incisors, (3) ankyloglossia (4) collapsed lower lip due to vertical bone loss caused by agenesis of the permanent mandibular anterior teeth.

The stages of treatment proposed for the patient were:

1. Insertion of a customized, functional, esthetic, fixed mandibular appliance, with acrylic teeth in the anterior region and bands in the permanent first molars—at 9 years, 7 months of age (Figure 3A-C).
2. Reduction in the proclination of the maxillary central incisors.
3. Surgical intervention for the correction of ankyloglossia.
4. Monitoring of the replacement of the primary teeth by their permanent successors.
5. Implants in the mandibular incisor area will be placed after alveolar growth has ceased.

The patient has currently been given a fixed appliance with banding on mandibular permanent molars and acrylic teeth in the mandibular anterior region (figure 7 and 8). There was significant improvement in esthetics and facial profile of the patient (figure 9 and 10). The patient was referred to a speech pathologist, she concluded that the patient did not have any speech pathology and the patient's parents didn't want to undergo surgery so surgical intervention for the treatment of ankyloglossia was abandoned.



Figure 7: Appliance in position front view

DISCUSSION:

The etiology of congenital absence of teeth is believed to be rooted in heredity or developmental anomalies.¹⁻³



Figure 8: Mandibular view



Figure 9: Front profile: note improvement



Figure 10: Lateral profile

Although oligodontia is genetic condition, the presence of external and internal factors, such as X-ray therapy, particular medications, infectious diseases, traumas, endocrine and intrauterine disorders, cannot be excluded. Familial tooth agenesis can occur as an isolated anomaly or as part of a genetic syndrome and is transmitted as an autosomal dominant, recessive, or X-linked condition.^{1,7} Studies have shown that MSX1

and PAX9 genes play a role in early tooth development.⁹ Treatment options for tooth agenesis should be customized for each patient, considering age, esthetics, and the need for rehabilitation.¹⁶ In the present case, the use of a fixed lingual arch-type appliance with acrylic teeth in place of the missing permanent teeth was considered suitable for maintaining the space required for future rehabilitation and for restoring the patient's smile. Given the patient's age, it was necessary to use a functional appliance that preserved the mandibular space until final prosthetic rehabilitation could be performed without interfering with the growth and development of the involved craniofacial structures.¹⁷ Frenectomy was not performed for the treatment of ankyloglossia because it didn't seem to cause any disability to the patient. In the maxillary arch, considering the patient's age, the proposed treatment was to reduce the proclination of the teeth to establish a better incisor relationship and to eliminate lip trap. Simultaneously, the vertical dimension could be improved. The next stage will consist of completion of the treatment with monitoring the eruption of remaining permanent teeth and finally prosthodontic rehabilitation with the help of implants when the age of the patient warrants.

CONCLUSION: Dentists should evaluate each case of tooth agenesis individually & over the long-term to prioritize the concerns of the patients and their parents while striving to achieve an esthetically and functionally favorable outcome taking into consideration the patient's growth and development.

REFERENCES:

1. Dhanrajani PJ. Hypodontia: etiology, clinical features, and management. *Quintessence Int* 2002;33:294-302.
2. Gorlin RJ, Cohen M Jr, Leven L, eds. *Syndromes of the head and neck*. 3rd ed. New York: Oxford University Press, 1990.
3. Polder BJ, Van't Hof MA, Van der Linden FP, Kuijpers-Jagtman AM. A meta-analysis of the prevalence of dental agenesis of permanent teeth. *Community Dent Oral Epidemiol* 2004;32:217-226.
4. Jepson NJ, Nohl FS, Carter NE, Gillgrass TJ, Meechan JG, Hobson RS, Nunn JH. The interdisciplinary management of hypodontia: restorative dentistry. *Br Dent J* 2003;194:299-304.
5. Schalk-van der Weide Y, Beemer FA, Faber JA, Bosman F. Symptomatology of patients with oligodontia. *J Oral Rehabil* 1994;21:247-261.
6. Hall RK. Congenitally missing teeth-a diagnostic feature in many syndromes of the head and neck. *Int Ass Dent Child* 1983;14:69-75.
7. De Coster PJ, Marks LA, Martens LC, Huysseune A. Dental agenesis: Genetic and clinical perspectives. *J Oral Pathol Med* 2009;38:1-17.
8. Worsaae N, Jensen BN, Holm B, Holsko J. Treatment of severe hypodontia-oligodontia: An interdisciplinary concept. *Int J Oral Maxillofac Surg* 2007;36:473-80.
9. Singer SL, Henry PJ, Lander ID. A treatment planning classification for oligodontia. *Int J Prosthodont* 2010;23:99-106.
10. Tsai PF, Chiou HR, Tseng CC. Oligodontia: A case report. *Quintessence Int* 1998;29:191-93.
11. Haavikko K. Hypodontia of permanent teeth. An orthopantomographic study. *Suom Hammaslaak Toim* 1971;67:219-25.
12. Pilo R, Kaffe I, Amir E, and Sarnat H, "Diagnosis of developmental dental anomalies using panoramic radiographs," *ASDC J Dent Child* 1987; 54:267-272.
13. Hintze H and Wenzel A. Longitudinal study of accuracy of clinical examination for detection of permanent tooth aplasia. *Community Dent Oral Epidemiol* 1990;18:256-259.
14. Derbanne MA, Sitbon MC, Landru MM, Naveau A. Case report: early prosthetic treatment in children with ectodermal dysplasia. *Eur Arch Paediatr Dent* 2010;11:301-305.
15. Kirzioğlu Z, KöselerSentut T, OzayErtürk MS, Karayilmaz H. Clinical features of hypodontia and associated dental anomalies: a retrospective study. *Oral Disease* 2005;11:399-404.
16. Fiorentino G, Vecchione P. Multiple congenitally missing teeth: Treatment outcome with autologous transplantation and orthodontic space closure. *Am J Orthod Dentofacial Orthop* 2007;132:693-703.
17. Tulunoğlu O, Cinar C, Bal C, Bal BT. Two-year study of alternative conservative treatment modalities for early anterior permanent tooth loss. *NY State Dent J* 2010;76:27-30.

Source of support: Nil

Conflict of Interest: None declared