PACIFIC JOURNAL OF MEDICAL SCIENCES

{Formerly: Medical Sciences Bulletin}

ISSN: 2072 – 1625

Pac. J. Med. Sci. (PJMS)

www.pacjmedsci.com. Emails: pacjmedsci@gmail.com; pacjmedsci1625@gmail.com

CASE REPORT:

A RARE CASE OF NONSYNDROMIC OLIGODONTIA WITH ANKYLOGLOSSIA

***Reshma Suvarna, Prasanna Kumar Rao, Raghavendra Kini, Devika Shetty and Vidya Holla**

Department of Oral Medicine and Radiology AJ Institute of Dental Sciences, Kuntikana, NH- Mangaluru Karnataka, India

***Correspondence author**: itsreshma_11@yahoo.co.in

Running Title: Oligodontia associated with ankyloglossia

CASE REPORT:

A RARE CASE OF NONSYNDROMIC OLIGODONTIA WITH ANKYLOGLOSSIA

***Reshma Suvarna, Prasanna Kumar Rao, Raghavendra Kini, Devika Shetty and Vidya Holla**

Department of Oral Medicine and Radiology AJ Institute of Dental Sciences, Kuntikana, NH- Mangaluru Karnataka, India

***Correspondence author**: itsreshma_11@yahoo.co.in

Running Title: Oligodontia associated with ankyloglossia

ABSTRACT:
Agenesis of teeth and ankyloglossia are common human developmental anomalies. Terms like Oligodontia, Anodontia and Hypodontia are used to describe agenesis of teeth. Oligodontia is a rare condition generally defined as agenesis of six or more teeth excluding the third molars. The condition is not frequently documented in Indian children. There is no much documentation on oligodontia with ankylogossia. Ankyloglossia can adversely affect the development of the surrounding structures including the alveolar process, teeth and can impair functions such as mastication, speech, and swallowing. The present article reports a rare case of non-syndromic oligodontia associated with ankyloglossia in an 8-year old male patient. Oligodontia is a relatively rare condition affecting 0.1-0.2% of the population. Our present case is even rare because of its association with ankyloglossia.

Keywords: Oligodontia, Ankyloglossia, Nonsyndromic, Agenesis.
Submitted in August, accepted in November 2017

INTRODUCTION:

One of the most common developmental anomalies seen in the permanent dentition is the agenesis of one or more teeth [1,2]. It is classified according to the number of missing permanent teeth excluding the third molars [3]. Hypodontia is the congenital agenesis of 5 or fewer permanent teeth; oligodontia is the congenital agenesis of 6 or more permanent teeth, and anodontia is the congenital agenesis of all deciduous and/or permanent teeth [1].

Hypodontia has a prevalence of 1.6% to 9.6% in the permanent dentition, excluding agenesis of the third molars. Oligodontia has a population prevalence of 0.3% in the permanent dentition. Oligodontia occurs more frequently in girls at a

ratio of 3:2 [2,4]. Agenesis of only the third molars has a prevalence of 9% to 37%. In the deciduous dentition, hypodontia occurs less often (0.1%-0.9%) and has no significant sex distribution [2, 4]. The mandibular second premolar is the most common missing tooth following third molar, followed by the maxillary lateral incisor and the maxillary second premolar [2, 5].

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] Oligodontia may also be a part of a genetic syndrome, as a non-syndromic isolated familial trait, as an infrequent finding or as an isolated condition that has been linked to mutations of the MSX1and m PAX9 [6].

The absence of teeth in young patients can cause esthetic, functional and psychological problems particularly if the teeth of the anterior region are involved.[2]

In this paper, we report a rare case of non-syndromic oligodontia with agenesis of ten permanent teeth excluding the third molars in a 8-year old patient along with the presence of ankyloglossia. There appears to be some relationship between ankyloglossia and agenesis of mandibular anteriors.

CASE REPORT:

An 8-year medically fit male patient, accompanied by her parents, reported to the department of Oral Medicine and Radiology with the chief complaint of missing lower front teeth since birth. He had no history of trauma and tooth extractions, without any family history. Further history revealed no teeth ever erupted in this region of the jaw, which suggests that the deciduous incisors were congenitally missing. Extraoral examination revealed, convex facial profile, marked labiomental fold with lip trap and reduced lower facial height. No facial asymmetry was detected. Intraoral clinical examination revealed absence of maxillary lateral incisors and primary maxillary first molars (Figure A) and mandibular central and lateral incisors and the tip of the tongue was attached to the lower alveolar ridge (Figure B). Ankyloglossia which was classified as Class IV by utilizing Kotlow's assessment and was not able to protrude the tongue and his speech was affected. The patient was in the mixed dentition stage. Panoramic radiograph was advised (Figure C). It revealed normal bilateral condyles and coronoid processes with its associated structures. Absence of tooth germs of the permanent teeth in relation to 12, 13, 22, 23, 31, 32, 41, 42, 43, and 44. The remaining permanent teeth were developing normally. Based on the clinical and radiological examinations, the diagnosis of non-

syndromic oligodontia was made. The condition was explained to the parents and patient and treatment plan was discussed. The patient was referred to the department of pedodontics for management of the oligodontia along with surgical intervention for the correction of ankyloglossia.

Fig A: Reveals missing lateral incisors and deciduous first molar

Fig B: Shows congenitally missing lower incisors in association with ankyloglossia

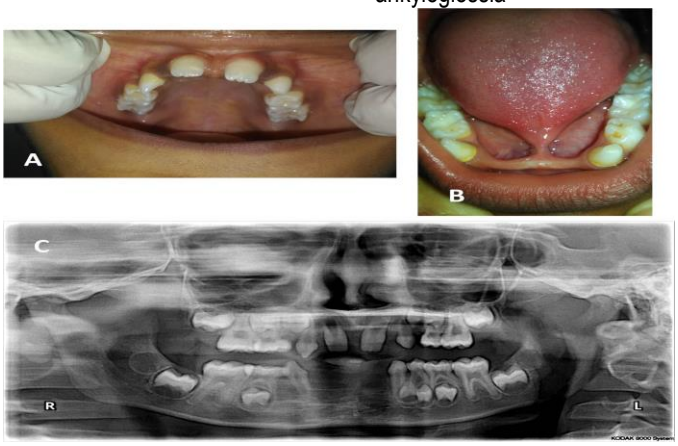

Fig C: OPG shows congenitally missing 12, 13, 22, 23, 31, 32, 41, 42, 43, and 44

DISCUSSION:

Tooth agenesis, the congenital absence of one or more permanent teeth, is a common human anomaly. A tooth is defined to be congenitally missing if it has not erupted in the oral cavity and is not visible in a radiograph [3]. The present case of congenitally missing 10 permanent teeth without any systemic disorders is suggested as isolated or non-syndromic oligodontia. Oligodontia is a rare anomaly, affecting approximately 0.1 to 0.3% of the population [3].

Hypodontia is an anomaly that may result in dental malpositioning, periodontal damage, lack of development of maxillary and mandibular bone height and has significant psychological, aesthetic and functional consequences.[2] The biologic basis for the congenital absence of permanent teeth is partially explained by the failure of the lingual or distal proliferation of the tooth bud cells from the dental lamina.[6]

The etiology of congenital absence of teeth is believed to be rooted in heredity or developmental anomalies.[1,7] Even though

oligodontia is genetic condition, factors such as X-ray therapy, certain medications, infectious diseases, traumas, endocrine and intrauterine disorders, may lead to oligodontia.[1]

Mutation in the transcription factors MSX1 and PAX9 have been identified in families. The factors were demonstrated to be associated with isolated non-syndromic oligodontia.[6] Recent studies have shown that mutation in EDA gene could result in non- syndromic oligodontia.[6] These isolated forms may be sporadic or familial. Familial tooth agenesis can be the result of single dominant gene defect, a recessive or X-linked.[6] Familial relationship suggests that the genes are important but in our present case family history was negative.[3] Oligodontia can occur in association with various genetic syndromes, such as ectodermal dysplasia, Down syndrome, Rieger syndrome , Van der Woude syndrome, Cleft lip palate ectodermal dysplasia syndrome, Oral facial digital syndrome type I, Hemifacial microsomia and others.[2] When oligodontia is associated with a syndrome there may be abnormalities of the skin, nails, eyes, ears and skeleton, which was not seen in the present case.

Oligodontia can be associated with malformations of crown, enamel hypopasia, failure of eruption, microdontia, macrodontia, and germination.[8] In our present case a rare association of oligodontia and ankyloglossia was seen. An extensive search of the current literature indicated that this association has not been reported previously in literature.

Ankyloglossia is defined as “a condition, in which the tip of the tongue cannot be protruded beyond the lower incisor teeth because of short frenulum linguae, often containing scar tissue”. In the present case, fusion of the tip of the tongue was attached to the lower alveolar ridge to the lower lip and missing of the deciduous as well as permanent teeth suggested that there has been a deviation from the normal development in the region of fusion of the first brachial arches. On the review of literature, various classifications of ankyloglossia have been proposed. [9] Thus, our present case was classified as Class IV by utilizing Kotlow's assessment. Frenectomy is the choice of treatment for ankyloglossia.

Panoramic radiography together with clinical examination of the oligodontia is recommended for the detection and confirmation of dental development and performing the diagnosis. [1] Treatment approach has to be case specific and depends on condition of primary predecessor, number of missing teeth, status of occlusion and patient and parent’s preferences. Options include orthodontic therapy, implants, removal partial prosthesis, fixed prosthesis, over dentures and indicated depending on the type of condition. Frenectomy was indicated for ankyloglossia Treatment; it does not only improve speech and

chewing function but also has psychological implications that may greatly help in regaining self-confidence [6].

CONCLUSION:

Complete understanding of rare anomalies like oligodontia and ankyloglossia may be enhanced by reporting of such cases. Such cases should be checked carefully for the presence of any syndromes and should be managed accordingly. Management of such patients generally requires a multidisciplinary approach. Treatment improves speech and chewing function and also has psychological implications that may have a huge impact on self-confidence.

REFERENCES:

1. 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.
2. Mallayya H C. Nonsyndromic Oligodontia: A Rare Case Report. AOSR 2012; 2 (2):103-107.
3. Bajaj P, Sabharwal R, Joshi S. Nonsyndromic Oligodontia: A Rare Case Report With Review Of Literature. Dental Journal of Advance Studies 2014; 2 (2):109-12.
4. Brook AH, Elcock C, Al-Sharood MH, McKeown HF, Khalaf K, Smith RN. Further Studies of a Model for the Etiology of Anomalies of Tooth Number and Size in Humans. Connect Tissue Res 2002;43:289-295.
5. Guruprasad R, Nair PP, Hegde K, Singh M: Nonsyndromic Oligodontia- a case report. IJDA 2011;3:450-453.
6. Hussein M A, Watted N, Zere E. Nonsyndromic Oligodontia in Permanent Dentition: Three Rare Cases. IOSR Journal of Dental and Medical Sciences 2015;14(12):79-83.
7. 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.
8. P, John JB, Priya G, Elango I, S. Familial nonsyndromic oligodontia. Contemp Clin Dent 2012;3:188-90.
9. Chandrashekar L, Kashinath K R, Setty SLabial Ankyloglossia Associated with Oligodontia: A Case Report. Journal of Dentistry, Tehran University of Medical Sciences 2014;11(4):481-4.