



Full Mouth Rehabilitation of a patient with Complete Anodontia: A Case Report

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Abstract

This article presents a case report of a 16 years old female patient who complained of retained deciduous teeth and unpleasant aesthetics. The Panoramic radiograph revealed complete absence of the successor teeth. The patient also presented with hyperkeratosis of toes and finger tips.

Key words Anodontia; hyperkeratosis; MTA

Introduction

Anodontia is defined by the National Organization of Rare Disorders as the congenital absence of all teeth in the primary dentition and/or the permanent dentition and it is a very rare condition. It is also known as dental agenesis. This congenital absence of teeth results



from the aplasia of the dental lamina due to either genetic or environmental factorsⁱ. The genetic aetiology was confirmed by studies on monozygotic twins. The environmental factors include infection, drugs, trauma, etc. Patients with anodontia inherit it from their parents in an autosomal recessive pattern. The most supported etiological theory suggests a polygenic type of inheritance, with epistatic genes and environmental factors exerting certain impact on the phenotypic expression of the genes involved, which then can disrupt the tooth germ during the initial stages of formation, i.e., the initiation and proliferation.ⁱⁱ The exact genetic mechanism is not recognised. Distinct mechanisms might as well account for missing of each tooth. Missing teeth are frequently seen as a part of a syndrome or as an isolated trait.ⁱⁱⁱ

In addition to an unesthetic appearance, patients with missing teeth may experience malocclusion, periodontal destruction, inadequate alveolar bone growth, reduced chewing capability, unintelligible pronunciation and several other problems. Due to deficient growth of the alveolar ridges, the vertical dimension of the face is reduced, the vermilion border of the lip recedes, leading to a senile appearance. Not only the physical aspects are disturbed, but the impact of anodontia on the psychological and psychosocial states of the individual are also traumatised. Considering all of these factors, the overall development of the child is hampered and therefore early full mouth rehabilitation is the best treatment plan in such cases.

The aim of this paper was to report a case of a 16-year-old patient with Anodontia and to discuss the approach in the overall management and the treatment of this condition..

Case description

A 16 years old female was brought into the Department of Pediatric and Preventive Dentistry with the chief complaint of retained deciduous teeth and unpleasant aesthetics. The patient was a well developed and well nourished female. The patient had a symmetrical face, everted lower lips and a prominent chin. (Fig 1)

On procuring the family history, it was found that the patient had two siblings wherein one sibling had the same condition. The parents had a consanguineous marriage. No other relevant family medical history was discovered.

The intra-oral examination revealed presence of deciduous dentition in the maxillary and mandibular arches except for the primary maxillary lateral incisors with normal mucosa and periodontal tissue.

Along with this, the patient presented with hyperkeratosis of finger tips and toes.

The panoramic radiograph divulged that all the permanent successor teeth were congenitally missing.





Fig 1. Well nourished, well developed 16 years old female. Symmetrical face and prominent chin with everted lip.



Fig 2. presence of deciduous dentition in maxillary and mandibular arches



Fig 3. Normal mucosal and periodontal tissue



Fig 4. Congenital absence of maxillary lateral incisors

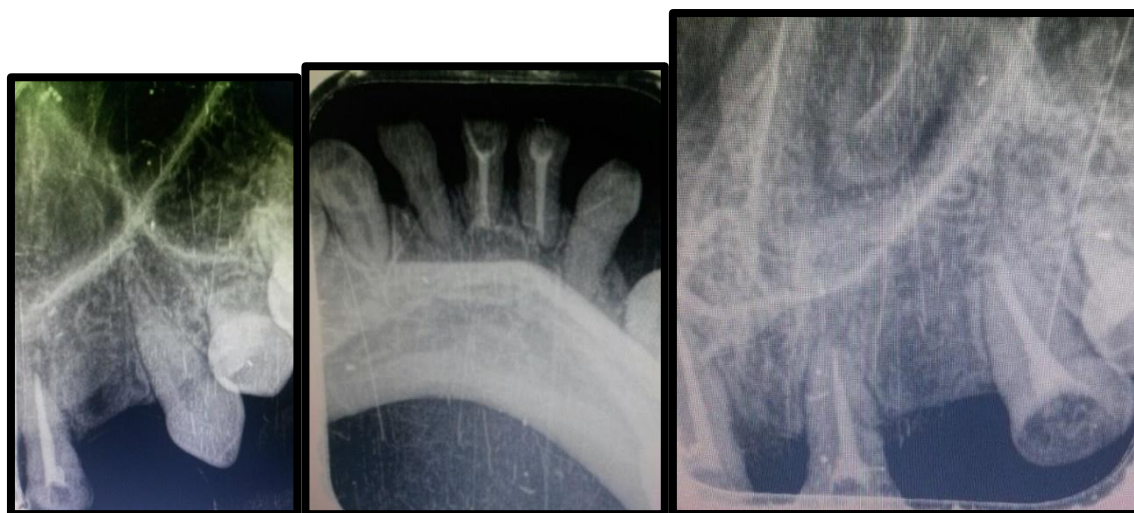


Fig 5. Hyperkeratosis of finger tips and toes



Fig 6. Panoramic radiograph showing congenital absence of permanent successors

A treatment plan was devised to preserve the primary teeth in the oral cavity for form and function. Pulpectomy was planned with 51, 53, 61, 71 and 81. Obturation was done with Mineral Trioxide Aggregate (MTA). Parent and patient motivation was done for maintenance of oral hygiene.



7. IOPA showing obturation with MTA obturation

Discussion

The evidence in literature for presence of deciduous teeth and congenital complete absence of permanent teeth is very limited. Oral rehabilitation of young patients with anodontia depends upon the age, number, condition of present teeth, and the stage of growth of the patient. Anodontia adversely affects the physiology as the absence of teeth is accompanied by vestigial ridge; therefore, restoring function and appearance are more perplexing than usual. It affects the psyche of the child as young patients realize that they are different from other children, hence confidence boost is also one of the primary objectives of the treatment plan.

Anodontia can appear in correlation with other genetic disorders as part of a documented clinical syndrome, or as a non-syndromic, hereditary form, which occurs as a solitary trait; shows a wide phenotypic heterogeneity, appears either intermittently or in a familial fashion within a family pedigree.^{iv} For a more holistic evaluation of the patient and especially in children for charting out the suitable management of dental anomalies, correlation between gene mutations and teeth agenesis is important for genetic counselling. PAX9 gene has been identified as a key controlling cause during the odontogenic process with its expression found unambiguously at the potential sites of all teeth prior to there being any morphological signs of odontogenesis.^v A general role for MSX1 is in the development of ectodermal derivatives as it is strongly expressed in the dental mesenchyme. The link between the gene and tooth agenesis was primarily found in a Finnish family with a predisposition for colorectal cancer.^{vi} The mutations of AXIN2 - Arg656Stop and 1994-1995insG lead to decreased AXIN2 function and most probably represent loss of function mutations that cause activation of Wnt signalling. AXIN2 was thought to be a strong contender gene for several reasons: Its position within this particular chromosomal region, a previously identified association with colorectal carcinoma and the fact that AXIN2 is also a known regulator of the Wnt signalling pathway. The Wnt family of secreted proteins forms part of a large family of signalling molecules that have a wide-ranging role during embryonic development and demonstrate regionally restricted expression in the tooth. The mode of transmission of hypodontia due to defects in the AXIN2 gene has not been definitively confirmed, and it has been seen that individuals with a non-sense mutation in AXIN2 display a mixed pattern of dental agenesis.

Hypodontia features in a number of other syndromes such as Rieger's syndrome, Oculofacial-cardio-dental syndrome, Incontinentia pigmenti, Pierre Robin sequence, Fried syndrome, Book syndrome, Down's syndrome, Wolf-Hirschhorn syndrome, Kabuki syndrome, Diastrophic dysplasia (DTD), Hemifacial microsomia and Recessive incisor hypodontia (RIH).^{vii}

Early rehabilitation is critical considering that the establishment of lifelong dietary patterns occurs during childhood.

Hyperkeratosis is a condition characterised by marked thickening of the outer layer of skin which is made up of keratin. It has been reported in a myriad of cases that hyperkeratosis accompanies anodontia.^{viii}

In this case, as the main goal was preservation of primary teeth, especially beyond their age of exfoliation, the protection of dental pulp was mandatory. Treatments that preserve tooth vitality are identified as vital pulp therapies. The aim of vital pulp therapy is to protect the pulp vitality, the tooth function and its supporting tissues in the case of dental caries, traumatic injuries and other conditions [AAPD, 2014].

MTA is a mineral material that when mixed with water results in a hydrated calcium silicate gel containing calcium hydroxide. It is also very alkaline and promotes tissue regeneration when placed in contact with the pulp or periradicular tissues.^{ix} It is biocompatible, non-toxic and non-resorbable and leads to minimal leakage around the margins. Valois(2004)



reported excellent marginal adaptation and sealing ability. Budig(2008) stated that the set cement has no signs of solubility, but increases when excess H₂O is added while mixing and cementogenesis is due to calcium hydroxide by the reaction of set MTA with water. Kettering(1995) identified MTA as a highly biocompatible material. MTA induces reparative dentin formation at a greater rate and has a superior structural integrity compared to calcium hydroxide. Various animal studies have constantly demonstrated that MTA induces dentin bridge formation that is thicker with lesser defects and side effects.^x

In this case, prosthetic rehabilitation will be required in the future for establishing accurate vertical dimension of the face and to restore and maintain the function and aesthetics. Since alveolar bone development is dependent on the presence of teeth, children with oligodontia or anodontia have atrophy of the alveolar bone and consequently little or no bone ridge to support dentures. Conventional removable complete or partial dentures are usually the treatment of choice for these patients.^{xi} Implant supported prosthesis depending upon the condition of the alveolar bone can also be considered. The effect of the future prosthetic management of the patient would be evaluated in long-term follow up.

Conclusion

The pulpectomy of the affected deciduous teeth in this case followed by obturation with MTA was successful for the preservation of deciduous teeth in the oral cavity for at least some time. The planning and implementation of prosthetic rehabilitation in the future will need a long-term treatment plan and follow up.

References

1. Ohno K, Ohmori I. Anodontia with hypohidrotic ectodermal dysplasia in a young female: a case report. *Pediatr Dent*. 2000;22(1):49-52.
2. Rakhshan V. Congenitally missing teeth (hypodontia): A review of the literature concerning the etiology, prevalence, risk factors, patterns and treatment. *Dent Res J (Isfahan)*. 2015;12(1):1-13.
3. Steijlen PM, Neumann HA, der Kinderen DJ, Smeets DF, van der Kerkhof PC, Happle R. Congenital atrichia, palmoplantar hyperkeratosis, mental retardation, and early loss of teeth in four siblings: a new syndrome? *J Am Acad Dermatol*. 1994;30(5 Pt 2):893-8.
4. Chhabra N, Goswami M, Chhabra A. Genetic basis of dental agenesis - molecular genetics patterning clinical dentistry. *Med Oral Patol Oral Cir Bucal*. 2014 1;19 (2):e112-9.
5. Kapadia H, Mues G, D'Souza R. Genes affecting tooth morphogenesis. *Orthod Craniofac Res*. 2007;10:237-44.



6. Jowett AK, Vainio S, Ferguson MW, Sharpe PT, Thesleff I. Epithelial-mesenchymal interactions are required for *msx1* and *msx2* gene expression in the developing murine molar tooth. *Development*. 1993;117:461-70.
7. Ferrante MI, Giorgio G, Feather SA, Bulfone A, Wright V, Ghiani M, et al. Identification of the gene for oral-facial-digital type I syndrome. *Am J Hum Genet*. 2001;68:569-76.
8. Bani M, Tezkirecioglu AM, Akal N, Tuzuner T. Ectodermal dysplasia with anodontia: a report of two cases. *Eur J Dent*. 2010 Apr;4(2):215-22. PMID: 20396456; PMCID: PMC2853819.
9. Cervino G, Laino L, D'Amico C, Russo D, Nucci L, Amoroso G, Gorassini F, Tepedino M, Terranova A, Gambino D, Mastroieni R, Tözüm MD, Fiorillo L. Mineral Trioxide Aggregate Applications in Endodontics: A Review. *Eur J Dent*. 2020;14(4):683-691.
10. Colombo M, Poggio C, Dagna A et al. Biological and physico-chemical properties of new root canal sealers. *J Clin Exp Dent*. 2018;10(02):e120–e126.
11. Teixeira Marques NC, Gurgel CV, Fernandes AP, Lima MC, Machado MA, Soares S, Oliveira TM. Prosthetic rehabilitation in children: an alternative clinical technique. *Case Rep Dent*. 2013;2013:512951.

