

HYPODONTIA: CLINICAL MANAGEMENT AND TREATMENT OPTIONS - CASE REPORT

ХИПОДОНЦИЈА: КЛИНИЧКИ МЕНАЏМЕНТ И ТЕРАПЕВТСКИ МОЖНОСТИ - ПРИКАЗ НА СЛУЧАЈ

Srbinska D.¹, Mijoska A.², Trpevska V.¹

¹Clinic of Orthodontics, University Dental Clinical Centre "St. Panteleimon", Skopje, Republic of North Macedonia, ²Department of Prosthodontics, Faculty of Dentistry - Skopje, Ss. Cyril and Methodius University in Skopje, Republic of North Macedonia

Abstract

Hypodontia is a condition used to describe the developmental absence of one or more teeth in primary or in permanent dentition, excluding the third molars. The tooth most commonly found to be missing is the third molar. It can be stated that a tooth is congenitally missing when it is clinically and radiographically (orthopantomogram) missing in the dental arch. Hypodontia can occur with or without syndrome. This developmental dental anomaly is the most common one and can be a challenge to manage clinically. The aetiology of hypodontia varies, so genetic, epigenetic and environmental may be contributory factors. Dental anomalies reported in literature associated with hypodontia include the following: microdontia, canine impaction, transposition, rotation of teeth, taurodontism and hypoplastic alveolar bone. Clinical management of hypodontia requires careful multidisciplinary planning. Congenital, bilateral absence of teeth is a very rare case in patients without syndrome. Orthodontic treatment of hypodontia depends on the area where the tooth is missing. Unerupted tooth might cause aesthetic and/or functional problems especially if it is situated in the maxillary anterior region. This article presents cases of hypodontia in early adolescent (agenesis of lateral incisor) and in adult period (bilateral absence of lateral incisors). **Key words:** hypodontia, aetiology, dental anomalies, orthopantomogram, orthodontic treatment.

Апстракт

Хиподонцијата е состојба која се користи при опишување на развоен недостаток на еден или повеќе заби во млечна или постојана дентиција, со исклучок на третите молари. Заб за кој е познато дека најчесто недостасува е третиот молар. Вроден недостаток на некој заб се смета онаа состојба кога и со клинички и со рендгенолошки преглед (ортопантомограм) одреден заб недостасува во денталниот лак. Хиподонцијата може да се јави во склоп на синдром или без појава на синдром. Оваа развојна дентална аномалија е многу честа појава и претставува предизвик во клиничкото менаџирање на истата. Етиологијата варира и како причинители за нејзина појава се наведуваат генетски, епигенетски и надворешни фактори. Во литературата може да забележиме дека хиподонцијата, како појава, се поврзува со одредени дентални аномалии, како што се: микродонција, импакција на канини, транспозиција, ротација на забите, тауродонтизам и хипопластична алвеоларна коска. Клиничкото менаџирање на хиподонцијата бара мултидисциплинарен и внимателен пристап. Вроден, обостран недостаток на забите е многу редок случај кај пациенти без синдром. Ортодонскиот третман на хиподонцијата зависи од регијата каде што недостасуваат забите. Вродениот недостаток на заби во максиларната фронтална регија доведува како до естетски така и до функционален проблем. Овој приказ покажува случаи на хиподонција во ран адолесцентен период (недостаток на латерален инцизив), и кај возрасен пациент (обостран недостаток на латерални инцизиви). **Клучни зборови:** хиподонција, етиологија, дентални аномалии, ортопантомограм, ортодонски третман.

Introduction

Hypodontia is developmental absence of one or more teeth and it is the most common dental anomaly in humans, often representing a major clinical problem¹. Congenitally missing teeth are classified according to the number of missing teeth, except the third molars. The term hypodontia is used to define agenesis of one to six teeth excluding the third molars. Oligodontia is absence of more than six teeth (excluding the third molars), also known as severe hypodontia, and anodontia is the complete absence of teeth^{2,3}. The incidence of hypodontia in

permanent dentition varies from 2.6% to 11.3% in the overall population, while the incidence in primary dentition is rare, ranging from 0.08% to 1.55%. Hypodontia in primary dentition generally appears in the anterior region and often associates with missing the same permanent tooth⁴. Tooth agenesis affects the maxilla and the mandible with similar prevalence⁵.

Most cases of hypodontia have a polygenetic inheritance pattern. Genetic studies suggest both, genetic and environmental aetiology toward this anomaly⁶. Various experimental studies show that the genes control the number and region of missing teeth. Mutation of genes,

such as MSX1 and PAX9, have been implicated with hypodontia in few studies⁷⁻¹⁰. Environmental factors, such as trauma in the dental region (fractures), surgical procedures on the jaws, chemotherapy and radiation therapy (depending of the age of the patient and the dose), may also contribute toward hypodontia¹¹. Somatic diseases such as syphilis, scarlet fever and rickets are also associated with hypodontia, as well as nutritional disturbances during pregnancy or infancy. Hypodontia can occur as an isolated dental anomaly (associated with lesser craniofacial or dental anomalies) or as part of a syndrome (Ectodermal dysplasia, Down syndrome). It has been reported that non-syndromic hypodontia occurs in connection with other dental anomalies such as microdontia, canine impaction, taurodontism, transposition, rotation of teeth and hypoplastic alveolar bone. Microdontia (reduction in tooth size), as one of the most common dental anomalies, is associated with hypodontia of a maxillary lateral incisor on one side and a peg-shaped lateral incisor on the other side¹². There is also relationship between palatal impacted canines, transposition of the maxillary canine and first premolar in cases of maxillary lateral incisor agenesis¹³. Other researchers have found that if there is a unilateral maxillary lateral incisor or premolar agenesis, it is more likely that the corresponding teeth on the other side will be rotated¹⁴.

The absence of teeth may introduce several signs and symptoms such as reduction of chewing ability, malocclusions, difficulty pronouncing words, compromised aesthetics, periodontal damage and alveolar bone deficiency. These complications cause functional (posterior absence of teeth), as well as aesthetic limitation (anterior absence of teeth). Furthermore, hypodontia may extremely affect the patient's behavioural pattern, self-esteem and social life¹⁵⁻¹⁶.

The complexity of hypodontia treatment varies widely, therefore managing hypodontia is a big challenge and needs multidisciplinary specialist approach¹⁷⁻¹⁸. There are not usually any noticeable changes on the skeletal pattern in the mild types of hypodontia, but it may be possible to see changes in cases of severe hypodontia. Unfortunately, there is no established formal procedure to manage patients with hypodontia. Treatment is individual and might range from single restorations to surgery and multiple restorations. Various options and methods have been suggested, such as: closing the space orthodontically; redistributing or re-opening of space for prosthetic appliances (crowns, adhesive or conventional bridges, removable prosthesis) and dental implants¹⁹.

In general, treatment options depend on the age of the patient, the severity of hypodontia, the amount of space available, the type of malocclusion, the degree of inherent crowding, oral health, patient's motivation and opin-

ion, skeletal pattern, soft tissue profile, bone anatomy and position of vital structures, as well the economic status of the patient (finances)¹⁵. Cooperation between different specialties (general dental practitioners, orthodontists, paediatric dentists, prosthodontists, oral and maxillofacial surgeons), provides the best individual results for each patient²⁰.

Case report (1)

A 13 -year-old male with permanent dentition was brought to the Clinic of Orthodontics for orthodontic treatment. The patient's main complaint was aesthetic, he has a big space between the central incisors (Figure 1). The patient did not complaint of pain, there were no signs of infection and he had good oral hygiene. Intraoral clinic examination revealed hypodontia of the permanent lateral incisor on the right side of maxilla, ½ Angle Class III on right and left side, anterior crossbite and spacing in the frontal teeth in the maxilla. According to morphological analyses, narrow mandible and asymmetric maxillary dental arch due to the hypodontia was diagnosed. Furthermore, the patient had reduced motility of the tongue due to low frenal attachment (frenulum linguae) and there was high maxillary frenum attachment (Figure. 2). The panoramic radiograph confirmed congenitally



Figure 1. Intraoral view (occlusion of the patient)



Figure 2. Intraoral view (low frenal attachment)



Figure 3. Panoramic radiograph of the patient

missing lateral incisor and presence of three third molars (Figure 3).

Therapy plan

Clinical management of this patient was necessary, and the orthodontic treatment included few steps. Frenectomy procedure would aid in tongue mobility, correct the tongue rest posture and establish a proper swallowing pattern. The oral surgeon performed the frenectomy, and the orthodontist started the therapy with fixed appliance (Figure. 4). Closure of the empty space (maxillary midline diastema) and re-opening of space for lateral incisor will be done by mesialization of the right central incisor. This patient is young and therefore, providing some type of final solution to replace the missing tooth in the long term should be a priority. There are two options for final restorative replacement of the missing tooth, a crown made over a dental implant or a prosthetic bridge. Temporary solution will be some prosthetic removable appliance. Also, the aim of treatment will be correction of occlusion, Angle Class 3 malocclusion and anterior crossbite bite of incisors achieved by fixed orthodontic appliance.

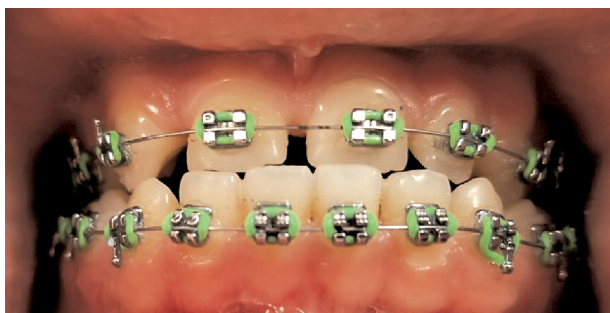


Figure 4. Therapy with fixed orthodontic appliance

Case report (2)

A 21-year-old female arrived at the Clinic of Orthodontics with main complaint of being “unhappy about her smile”. Her family history was non-contributo-



Figure 5. Intraoral view (anterior posture of the tongue)



Figure 6. a) (rotated second premolar in right maxilla)
b) (presence of primary canine)

ry with congenitally missing teeth. Extra oral examination revealed a facial asymmetry, whereas intra oral clinical examination revealed bilateral absence of lateral maxillary incisors, presence of maxillary primary canine, rotation and crossbite of permanent second premolar (Figure. 5 and Figure. 6 a, b). The molar relationship was ½ Angle Class II, there was anterior open bite and the overjet was 4 mm. Due to the presence of primary right maxillary canine, dental arch asymmetry was diagnosed. Panoramic radiograph confirms hypodontia of both later-

al incisors in the maxilla (Figure. 7). There is absence of third molars in the mandible. Furthermore, there was a poor oral habit (anterior posture of the tongue).

The aim of treatment of this clinical case include correction of the bad habit, levelling and coordinating the dental arches, correcting the centreline discrepancy, overjet and overbite, crossbite, achieving Angle Class I and retaining the corrected results.

Therapy plan

The stability of corrected relations after orthodontic treatment depends on regular tongue function. Therefore, based on clinical and radiographical examination, the correction of tongue posture with an individual myofunctional appliance was treatment priorit. Also, we recommended some oral myofunctional exercises. Appropriate use of this appliance and regular exercises (at least three times a day) give excellent results in correction of anterior tongue posture and facial asymmetry (Figure 8). The therapy will be continued with fixed orthodontic appliances that will correct the crossbite and rotation of the second premolar, the centreline, open bite, overjet and overbite. The final aim is to achieve “Happy smile”, by remodelling canine into lateral incisors by laminates or dental composites.

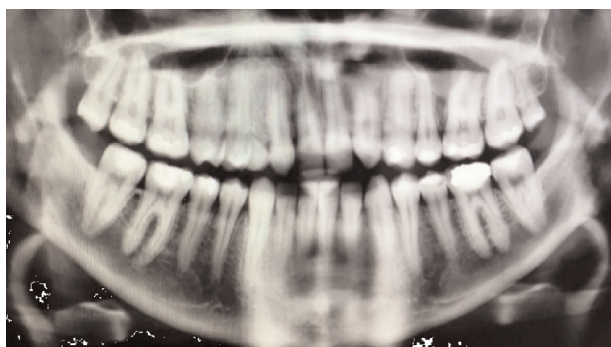


Figure 7. Panoramic radiograph of the patient



Figure 8. Myofunctional individual orthodontic appliance

Discussion

Diagnosis of dental anomalies is usually performed by paediatric dentists, as these professionals are the first to interact with children and adolescents²¹. The knowledge of odontogenesis is fundamental for understanding growth and developmental disorders affecting teeth. Hypodontia can be treated with different orthodontic modalities. The decision depends on various factors (patient's age, facial profile, severity of hypodontia, shape and size of the adjacent teeth, smile and gingival line)²². Presence of dental malocclusion, as well as poor oral habit, are very important for the final treatment plan. The orthodontic treatment in the first presented case ($\frac{1}{2}$ Angle Class III malocclusion) will be re-opening the space for lateral incisor after the frenectomy. The second presented patient ($\frac{1}{2}$ Angle Class II malocclusion), after correction of tongue posture, will be treated with orthodontic fixed appliances with camouflage treatment. The presented dental open bite and increased overjet is the result of incorrect rest tongue posture and lips posture²³⁻²⁴. The goal of orofacial myofunctional therapy is to correct oral rest posture of the tongue and lips which means, at rest, the lips should be closed without strain, the patient can breathe nasally, the tongue is resting against the mouth roof not touching the teeth.

These cases illustrate the need for a multidisciplinary team approach (orthodontic, dental practitioner, prosthetic and oral surgeon)²⁵. Correction of myofunctional dysfunction is priority for any malocclusion and stability of corrected results depends on this. According to most authors, there is no age limit to perform myofunctional therapy and the results depend on the patient's cooperation. Age is an important factor in determining the type of treatment for hypodontia cases. Most authors agreed that priority in orthodontic treatment is correction of poor oral habits, myofunctional disorders and the stability of achieved results depends directly on these factors.

Conclusion

Hypodontia presents a complex problem to dentists worldwide. Clinical management of hypodontia requires careful multidisciplinary planning and has financial implications. A number of procedures can be performed to fulfill the patient's wishes considering their age. The primary motivating factor for individuals seeking orthodontic treatment is aesthetics, whereas the best time for orthodontic treatment with agenesis is early adolescence, since it is a period when most of remaining developing permanent teeth are erupting and notable facial growth has happened.

Reference

1. De Coster PJ, Marks LA, Martens LC, Huysseune A. Dental agenesis: genetic and clinical perspectives. *J Oral Pathol Med.* 2009;38(1):1-17. [PubMed] [Google Scholar]
2. Hobkirk JA et al. Hypodontia: 2. The management of severe hypodontia, *Dental update* 1995;22:8-11.
3. Arte S. Phenotypic and genotypic features of familial Hypodontia. University of Helsinki. Thesis 2001
4. Khalaf K, et al. Prevalence of hypodontia and associated factors: a systemic review and met-a analysis. *Journal of orthodontics* 2014;41:299-316.
5. Polder BJ, Van't Hof MA, Van Der Linden FPGM, Kuijpers-Jagtman AM. A meta-analysis of the prevalence of dental agenesis of permanent teeth. *Community Dentistry and Oral Epidemiology* 2004;32(3):217-26.
6. Vastaridis H. The genetics of human tooth agenesis: new discoveries for understanding dental anomalies. *Am J Orthod Dentofacial Orthop* 2000;117:650-6.
7. Burzynski NJ, Escobar VH. Classification and genetics of numeric anomalies of dentition. *Birth Defects Orig Artic Ser* 1983;19:95-106.
8. Vastaridis H, Karimbux N, Guthua SW, Seidman JG, Seidman CE. A human MSX1 homeodomain missense mutation causes selective tooth agenesis. *Nat Genet* 1996;13:417-21
9. Stockton DW, Das P, Goldenberg M, D'Souza RN, Patel PL. Mutation of PAX9 is associated with oligodontia. *Nat Genet* 2000;24:18-9.
10. Van den Boogaard MJ, Dorland M, Beemer FA, van Amstel HK. MSX1 mutation is associated with orofacial clefting and tooth agenesis in humans. *Nat Genet* 2000;24:342-3.
11. Maguire A, Craft AW, Evans RG, Amineddine H, Kernahan J, Macleod RI, et al. The long-term effects of treatment on the dental condition of children surviving malignant disease. *Cancer* 1987; 60:2570-5.
12. McKeown HF, Robinson DL, Elcock C, Al-Sharood M, Brook AH. Tooth dimensions in hypodontia patients, their unaffected relatives and a control group measured by a new image analysis system. *Eur J Orthod* 2002;24:131-41.
13. Pirinen S, Arte S, Apajalahti S. Palatal displacement of canine is genetic and related to congenital absence of teeth. *J Dent Res* 1996;75:1742-46.
14. Lai PY, Seow WK. A controlled study of the association of various dental anomalies with hypodontia of permanent teeth. *Pediatr Dent* 1989;11:291-96.
15. Nasrin R. Sadaqah, Jawad Abu Tair. Management of patient with Hypodontia: Review of Literature and case report. *Open Journal of Stomatology* 2015;5:293-308.
16. Laing E, Cunningham SJ, Jones S, Moles D, Gill D. Psychosocial impact of hypodontia in children. *AJODO.* 2010;137(1):35-41.
17. Renato R.et al. A multidisciplinary treatment of congenitally missing maxillary lateral incisor: a 14-year follow-up case report. *J. Appl. Oral Sci* 2014;22.
18. Clare McNamara, Tim Foley, Catherine McNamara. Multidisciplinary Management of Hypodontia in Adolescents: Case report. *JCDA* 2006;72:740-746.
19. Steven Jones, John Hobkirk, Steven Bassi, et al. Counselling patients with Hypodontia. *Dental Update* 2008;35:344-352.
20. Hobkirk JA, Gill DS, Jones SP, Hemmings KW, Bassi GS, O'Donnell AL, et al. Hypodontia: A team approach to management (1 st ed). Chichester: Wiley-Blackwell 2011.
21. Moskowitz EM, Samson GS. Age-appropriate orthodontic treatment options: a joint responsibility for orthodontists and pediatric dentists. *Semin Orthod.* 2014;20(1):2-2. [Google Scholar]
22. Pinho T, Ciriaco C, Faber J, Lenza MA. Impact of dental asymmetries on the perception of smile aesthetics. *AJODO.* 2007;27(5):443-9.
23. Smithpeter J, Covell D., Jr Relapse of anterior open bites treated with orthodontic appliances with and without orofacial myofunctional therapy. *Am J Orthod Dentofacial Orthop.* 2010;137(5):605-614. doi: 10.1016/j.ajodo.2008.07.016.DOI: [PubMed] [Cross Ref] [Google Scholar]
24. Khemka S, Thosar N, Baliga S. Oral gymnastics - way to a harmonious dentition. *Int J Contemp Dent Med Rev.* 2015 [Google Scholar]
25. Carter NE, Gillgrass TJ, Hobson RS, et al. The interdisciplinary management of hypodontia: orthodontics. *British Dental Journal.* 2003;194(7):361-6.