

ŞİDDETLİ OLİGODONTİ İLE BİRLİKTE DAİMİ MAKSİLLER 1. MOLAR DIŞLERİN KONJENİTAL EKSİKLİĞİ: NADİR BİR OLGU SUNUMU

CONGENITALLY MISSING MAXILLARY FIRST PERMANENT MOLARS WITH SEVERE OLIGODONTIA: AN UNUSUAL CASE REPORT

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Özet

10 yaşında erkek hasta eksik diş şikayeti ile Ondokuz Mayıs Üniversitesi Oral ve Maksillofasial Radyoloji bölümüne başvurdu.

6 ya da daha fazla daimi dişin konjenital yokluğu oligodonti olarak adlandırılır. Oligodontinin genetik orijini tam olarak bilinmemektedir. Daimi maksiller birinci molar dişlerin konjenital eksikliği nadir bir durumdur. En yaygın eksikliği görülen dişler daimi maksiller lateral kesicilerdir, mandibular ikinci premolar dişler, maksiller ikinci premolar dişler ve mandibular santral kesiciler bunları izler. Özellikle bu dental anomali mandibulada görülür, daimi maksiller birinci molar dişlerin yokluğu sıklıkla ciddi ortodontik probleme sebep olabilir.

Sonuç olarak, oligodontinin erken teşhisi çok önemlidir ve tedavi multidisipliner yaklaşımla yapılabilir. Bu vakada daimi maksiller birinci molar dişlerin şiddetli oligodontiyle birlikte agenezisi sunulmuştur.

Anahtar Kelimeler: Oligodonti, multidisipliner, diş agenezisi.

Abstract

A 10 year-old male patient was referred to Department of Oral and Maxillofacial Radiology, Ondokuz Mayıs University, Samsun with a complaint of missing teeth.

The congenital absence of six or more permanent teeth is termed oligodontia. The genetic origin of oligodontia isn't precisely known. Congenitally missing maxillary first permanent molars is a rare condition. The most commonly missing teeth are maxillary permanent lateral incisors followed by the mandibular second premolars, maxillary second premolars and the mandibular central incisors. Especially this dental anomaly is seen in mandible, the absence of maxillary first permanent teeth may often cause serious orthodontic problem.

As a result, the early diagnosis of oligodontia is very important and the treatment may be performed in a multidisciplinary approach. We report a rare case of agenecies of maxillary first permanent molars with severe oligodontia.

Key words: Oligodontia, multidisciplinary, tooth agenesis.

Introduction

Congenital absence of teeth is most frequently seen as human dental anomalies. Affecting approximately %20 of the population in all over the world, this dental anomaly most commonly seen in third molars but also involves other teeth at varying frequencies (1,2). The congenital missing of the maxillary and mandibular permanent first molars have the least frequency (3). Congenitally missing maxillary first permanent molars accompanied by severe oligodontia suggests some hereditary

diseases such as Rieger's syndrome, hypohydrotic type of ectodermal dysplasia, incontinentia pigmenti, Hallerman- Streiff syndrome and premolare aplasia-hyperhydrosis- cavities prematura (PHC) syndrome (4). Tooth agenesis may be a part of medical or genetic syndrome. In fact, any syndromes or sistemic diseases are not seen in many patients who have congenitally missing teeth. The nonsyndromic form, hypodontia is absence of fewer than six congenitally missing permanent teeth and most commonly diagnosed. More severe rare nonsyndromic form is defined by oligodontia that greater than six missing permanent teeth is present (5). Predisposing factors of congenitally missing teeth are several. The first of these factors is the developmental anomaly during tooth-bud initiation and proliferation (6). Lesions of the jaws that occur during infancy, are also thought as suspicious factor. Even if the patient has

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radiation therapy at the early stage of tooth germ formation, this may cause the absence of one or more dental units (7). It was reported that hereditary factor caused one or more missing teeth in patients who have partial anadontia (8).

We aim to present a rare case of congenitally missing maxillary first permanent molars with severe oligodontia in a 10 year old male patient and the treatment options.

Case Report

A 10 year-old male patient was sent to our department for missing permanent teeth. The patient has no systemic disease. His medical history was non-contributory. In extraoral examination there was no abnormality. In intraoral examination, there was maxillary constriction, deep bite, posterior cross bite on the right side and median diastema (Figure 1,2a-b).



Figure 1. Pretreatment frontal view showing, deep bite, median diastema.



Figure 2a-b. Pretreatment sagittal view showing deep bite, posterior cross bite on the right side and median diastema.

Due to median diastema, an abnormal frenilum was seen. It exhibited excessive thickness, alveolar attachment between maxillary central incisors and large incisive papilla. Panoramic radiograph revealed anterior diastema, carious lesions in right and left

maxillary first primary molars, left mandibular first and second primary molars, right mandibular first primary molar and following congenital missing primary teeth: right and left maxillary second primary molars and following several congenital missing permanent teeth: right and left maxillary lateral incisors and canin teeth, right maxillary first premolar, right and left maxillary second premolars, first and second molars, right and left mandibular central incisors, right and left mandibular second premolars, first and second molars. So that, nonsyndromic severe oligodontia was diagnosed (Figure 3,4).



Figure 3. Pretreatment occlusal view of maxilla showing maxillary constriction with oligodontia.



Figure 4. Pretreatment occlusal view of mandible showing oligodontia.

The parents of the patient were informed of the condition. The occurrence of oligodontia, median diastema, deep bite and posterior cross bite, multidisciplinary approach was considered. The treatment planning that included two phases, was performed. Phase 1 involves orthodontic treatment and speech therapy, phase 2 involves prosthetic treatment. The primary phase should be performed immediately but the secondary phase is performed at older age when the cranio-facial growing of the patient finishes.

In phase 1 treatment, Maxillary arch width was expanded to correct posterior cross bite on the right side. It was aimed to correct the median diastema and deep bite and a retainer with denture teeth was made for replacing congenitally missing permanent teeth and providing function, phonation and esthetics of the patient. The patient was followed up till his adulthood to perform the secondary phase.

In phase 2 treatment, cranio-facial growing should be considered. When the patient have reached an age of 17-18 years, the treatment planning are evaluated again and definite treatment is determined such as orthognathic surgery, bone augmentations, dentures and implants. Due to the cranio-facial growing of our patient wasn't complete, temporary partial dentures were made in maxilla and mandible (Figure 5,6,7,8a-b, 9a-b).



Figure 5. Occlusal view of the mandibular removable partial denture.

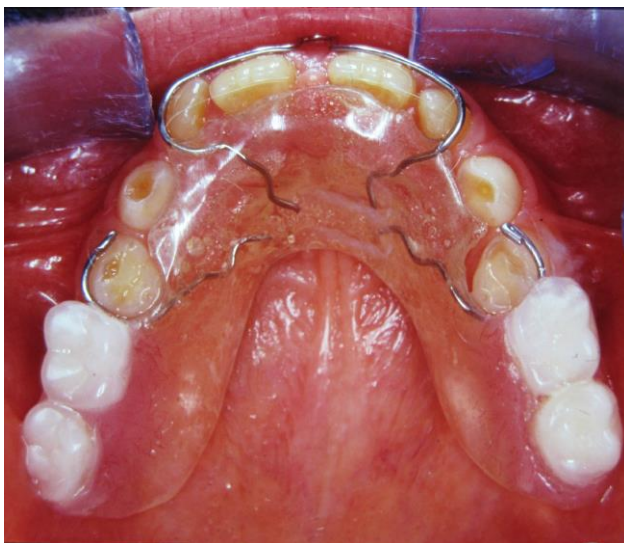


Figure 6. Occlusal view of the maxillary removable partial denture.



Figure 7. Frontal view of removable partial dentures.



Figure 8a-b. Maxillary removable partial denture.



Figure 9a-b. Mandibular removable partial denture.

The occlusion and adaptation of dentures were controlled. After the temporary dentures were inserted, when the final partial dentures could be made, the patient was informed in order to use them continuously for the success of orthodontic treatment until the age of skeletal maturity. Informations about how to use the dentures were given to the patient and his family and the patient was informed to return once a week for a month for controlling the adaptation of the dentures.

Discussion

There were rare isolated cases of oligodontia involving maxillary first permanent molars without abnormalities in the literature (9-11). In this case the patient had no other

associated abnormalities. Except third molars, the absence of permanent molar teeth is a rather rare occurrence (12). The first molars are very probably the most stable teeth because there were several studies (13-20) showing that these teeth are never absent. So our case report is extremely rare.

Hypodontia of primary dentition is rare and it was reported that the prevalence depends on various factors such as geographic location (0.1% to 0.9% bases on studies in Iceland, Scandinavia and Britain) and the study methods used (21-23). Reported incidence in Japanese population was higher with the rate of 5% (24). Idiopathically, missing primary molars are extremely rare condition (25,26). In this case, right and left maxillary primary second molars was absent. It was reported that the strong correlation between the agenesis of primary teeth and the agenesis of corresponding permanent teeth (27,28). Grahnen and Granath reported that %75 of cases with hypodontia in primary dentition had this anomaly in permanent dentition (25). Our case report partially supported this idea in maxillary arch. Together with agenesis of right and left maxillary second permanent premolars, missing right and left maxillary second primary molars were present in our case.

Muller et al. reported that congenitally missing permanent teeth are most frequently seen in girls than boys (29). Agurto Goya et al. also reported that the girls were affected almost twice as often as boys (30). In this case, our patient with severe oligodontia was a boy.

Kırzioğlu et al.(31) found more missing teeth in mandibular arch (50.3%) than maxillary arch (49.7%) but Silva Meza and Rasmusen found the predominance of maxillary arch as 55.2 and 52.1, respectively (32). Salama and Abdel-Megid also found that maxillary missing teeth (52%) were more frequently seen than mandibular missing teeth (48%) (33). In our case, in concordance with the study of Silva Meza and Rasmusen, Salama and Abdel-Megid, we found more missing teeth in maxillary arch than mandibular arch.

Agenesis of maxillary central incisors, maxillary and mandibular first molars and canines were reported to be very rare in the study of Kırzioğlu et al in concordance with the studies of Dhanrajani, Zarrinnia and Bassiouny (31,34,35). In our case, right and left maxillary canines, right and left maxillary first and second

molars, right and left mandibular first and second molars were absent.

Generally, agenesis of teeth in the permanent dentition should be diagnosed after the age of 6 years except third molar, the third molar is included after 10 years of age (36)

There are many factors in the treatment planning of oligodontia especially when several missing teeth is present and there is a greatest challenge for interdisciplinary team (37-39) So the multidisciplinary approach was planned involving orthodontics and prosthodontics to be sure adequate treatment. The orthodontic treatment was very important to establish the correct position of teeth (40) Prosthodontic treatment included these aims:

1. Provide masticatory function
2. Protect the position of adjacent natural teeth to prohibit unwanted movements such as migration or extrusion, inclination
3. Provide esthetics
4. Prevent social problems of the patient especially in adolescence
5. Replace the missing teeth without intervene in the growth of maxilla and mandible (40)

Implant wasn't indicated due to the age of patient. When the implant treatment is performed, it must be postponed until after puberty or after the growth of the child (41-43). However, atrophy of alveolar bone is another reason for implant contraindication. This condition is commonly seen in patients with oligodontia and results in decreased durability of implants (44,45). Therefore, we didn't think implant treatment for our patient. the jaw development of our patient wasn't completed so temporary removable partial denture was performed. This type of denture doesn't prevent the jaws development and eruption of permanent teeth (40).

This case represented severe oligodontia that has necessity for orthodontics and prosthodontics treatment and the importance of early diagnosis was determined. When the treatment completed, the patient had acquired the function, phonation and esthetics.

Conclusions

In conclusion, oligodontia is rare condition and is seen rarely in non-syndromic patients. The patients with oligodontia need

multidisciplinary approach so that they should be evaluated carefully. Early diagnosis of oligodontia has important role to prevent the complications of this anomaly and provide function, phonation and esthetics.

References

1. Online Mendelian Inheritance in Man. 1999 <http://www3.ncbi.nlm.nih.gov/omim/>
2. Shapiro SD, Farrington FH. Birth Defects Original Article Series 1983; 19 : 129-40.
3. Adsheed F 3rd. Common congenitally missing permanent teeth. Bull Phila City Dent Soc. 1970; 36: 14-6.
4. Schulze C. Developmental abnormalities of the teeth and jaws. In Thoma's Oral Pathology, Gorlin RI, Goldman HM. Eds. St. Louis, The C.V. Mosby Company. 1970: 96-183.
5. Frazier-Bowers SA, Scott MR, Cavender A, Mensah J, D'Souza RN. Connect Tissue Res. 2002; 43: 296-300.
6. Graber LW. Congenitally absent teeth: A review with emphasis on Inheritance Patterns. J Am Dent Assoc. 1978; 96: 266-75.
7. Stafne EC. Oral Roentgenographic Diagnosis. 3RD ed. W.B. Saunders Co. 1969; 14-41.
8. Stones HH. Oral and Dental Diseases. 3rd ed. E.&S. Livingstone LTD. 1954; 131-45.
9. Turner CH, Turner JL. Oligodontia: report of a case. Journal of Dentistry for Children 1977; Jan-Feb.: 49-51.
10. Neal RG, Lambdin LJ. Anodontia of a molar region. J. Of Pedodontics 1980; Winter: 179-81.
11. Jarvinen S, Vaataja P. Congenitally missing first permanent molars. J of Pedodontics 1981; Summer: 323-6.
12. Nieminen P, Arte S, Tanner D, Paulin L, Alaluusua S, Thesleff I et al. Identification of a nonsense mutation in the PAX9 gene in molar oligodontia. European Journal of Human Genetics 2001; 9: 743-6.
13. Nik-Hussein NN. Hypodontia in the permanent dentition: a study of its prevalence in Malaysian children. Aust Orthod J 1989; 11: 93-5.
14. Aasheim B, Ögaard B. Hypodontia in 9-year-old Norwegians related to need of orthodontic treatment. Scand J Dent Res 1993; 101: 257-60.
15. Okamoto O, Mori O, Morimoto M, Nakao N, Miyakawa E. A statistical and genetic study related to congenitally missing teeth. Shika Gakuho 1951; 5: 8-10,39-46,71-74 (in Japanese).
16. Ma C. Statistical observation of morphological and numerical teeth anomalies in the teeth of Japanese. Shikagaku Zasshi 1949; 6: 248-56 (in Japanese).
17. Terasaki T, Shiota K. Congenital absence of teeth. Nihon Koku Kagakkai Zasshi 1954; 3: 88-93 (in Japanese).
18. Yanagida I, Mori S. Statistical studies on numerical anomalies of teeth in children using orthopantomograms: congenitally hypodontia. Osaka Daigaku Shigaku Zasshi 1990; 35: 580-93 (in Japanese).
19. Ishizuka K, Sasaki T, Imai R, Nakamura N, Yoshida T, Anabuki M et al. Abnormalities of teeth which affects the orthodontic treatment. Nichidai Shigaku 1988; 62: 584-95 (in Japanese).
20. Loch S. Panoramic radiographic examination of 704 Danish children aged 9-10 years. Community Dent Oral Epidemiol 1980; 8: 375-80.
21. Moller P. Oral health survey of preschool children in Iceland. Acta Odontol Scand 1963; 21: 47-97.
22. Ravn JJ. Aplasia, supernumerary teeth and fused teeth in the primary dentition. An epidemiological study. Scand J Dent Res 1971; 79: 1-6.
23. Brook AH. Dentine anomalies of number, form and size: their prevalence in British School Children. J Int Assoc Dent Child 1974; 5: 37-53.
24. Saito TA. Genetic study on Degenerative anomalies of deciduous teeth. Jpn J Hum Genet 1959; 4: 27.
25. Grantham H, Granath LE. Numerical variations in primary dentition and their correlation with the permanent dentition. Odont Revy 1961; 12: 348-57.
26. Bennet CG, Ronk SL. Congenitally missing primary teeth: report of case. J Dent Child 1980; 47: 346-8.
27. Whittington BR, Durward CS. Survey of anomalies in primary teeth and their correlation with permanent dentition. New Zealand Dent J 1996; 92: 4-8.
28. 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 1993; 21: 162-4.
29. Muller TP, Hill IN, Peterson AC, Blayney JR. A survey of congenitally missing permanent teeth. J Am Dent Assoc 1970; 81: 101-7.
30. Goya HA, Tanaka S, Maeda T, Akimoto Y. An orthopantomographic study of hypodontia in permanent teeth of Japanese pediatric patients. Journal of Oral Science 2008; 50: 143-50.
31. Kırzioğlu Z, Şentürk Köşeler T, Ertürk Özyay MS, Karayılmaz H. Clinical features of hypodontia and associated dental anomalies: a retrospective study. Oral Diseases 2005; 11: 399-404.
32. Silva Meza R. Radiographic assessment of congenitally missing teeth in orthodontic patients. Int J Paediatr Dent 2003; 13: 112-6.
33. Salama FS, Abdel-megid FY. Hypodontia of primary and permanent teeth in a sample of Saudi children. Egypt Dent J 1994; 40: 625-32.
34. Dhanrajani PJ. Hypodontia: etiology, clinical features, and management. Quintessence Int 2002; 33: 294-302.
35. Zarrinnia K, Bassiouny MA. Combined aplasia of maxillary first molars and lateral incisors: a case report and management. J Clin Pediatr Dent 2003; 27: 127-32.
36. Arte S, Pirinen S. Hypodontia. Available online at www.orpha.net/data/patho/GB/uk-hypodontia.pdf
37. Hobson RS, Carter NE, Gillgrass TJ, Jepson NJ, Meechan JG, Nohl F, et al. The interdisciplinary management of hypodontia: the relationship between an interdisciplinary team and the general dental practitioner. Br Dent J 2003; 194: 479-82.
38. 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.
39. Bural C, Oztas E, Ozturk S, Bayraktar G. Multidisciplinary treatment of non-syndromic oligodontia. Eur J Dent 2012; 6: 218-26.
40. Gonçalves TMSV, Gonçalves LM, Sabino-Bezerra JR, Santos-Silva AR, José da Silva W, Garcia RCMR. Braz. Dent. J. 2013; 24: 174-8.
41. Locker D, Jokovic A, Prakash P, Tompson B. Oral health-related quality of life of children with oligodontia. Int J Paediatr Dent 2010; 20: 8-14.
42. Shilpa, Thomas AM, Joshi JL. Idiopathic oligodontia in primary dentition: case report and review of literature. J Clin Pediatr Dent 2007; 32: 65-7.
43. Artopoulou II, Martin JW, Suchko GD. Prosthodontic rehabilitation of a 10-year-old ectodermal dysplasia patient using provisional implants. Pediatr Dent 2009; 31: 52-7.
44. Finnema KJ, Raghoobar GM, Meijer HJ, Vissink A. Oral rehabilitation with dental implants in oligodontia patients. Int J Prosthodont 2005; 18: 203-9.
45. Mankani N, Chowdhary DR, Patil DB, E DN, Madalli DP. Dental implants in children and adolescents: A literature review. J Oral Implantol 2012 [Epub ahead of print. DOI: 10.1563/AAID-JOI-D-11-00186]