Journal of Morphology and Anatomy

Case Report Open Access

Bi-Maxillary Hyper-Hypodontia: A Unique Case Report with Review of Literature

Bharti C[1]*, Ratre RK[2], Ratre M[3], Jain S[2] and Singh B[4]

[1]*Department of Orthodontics, People's College of Dental Sciences and Research, Bhopal, MP, India*
[2]*Department of Orthodontics, Government College of Dentistry, Indore, MP, India*
[3]*Department of Periodontics, Government College of Dentistry, Indore, MP, India*
[4]*Ismile Believe Clinic, Indore, MP, India*

Abstract

The term "hypo-hyperdontia" is used to describe condition in which agenesis of some teeth are seen to occur simultaneously with supernumerary teeth within the same individual. Supernumerary teeth may resemble the permanent teeth sometimes but generally are of different shape and size. These teeth may remain embedded in the alveolar bone may cause disturbance to the developing teeth or can erupt into the oral cavity. Contrary to the presence of extra tooth in arch few complement of the teeth were missing in the arch indicating presence of concomitant hyper hypodontia. The erupted or unerupted tooth might cause aesthetic and/or functional problems especially if it is situated in the maxillary anterior region. Recently, many genetic studies have been conducted to explore these developmental anomalies. This case reports a unique case of presence of supplemental supernumerary maxillary central incisor with hypodontia of both lower central incisors, which is a rare finding and a literature review, is presented about hypo-hyperdontia in the dentition.

Keywords: Agenesis; Hyperdontia; Hypodontia

Introduction

Development of the tooth is a continuous process with a number of physiologic growth processes and various morphologic stages interplay to achieve the tooth's final form and structure. One such mixed rare numeric anomaly is concomitant hypo-hyperdontia (also known as oligo-pleiodontia) characterized by developmental absence of the teeth (hypodontia) and supernumerary teeth (hyperdontia) co-existing in the same individual [1]. It is the only event in which the developing organ must exit the confines of its bony crypt. Interference with the stage of initiation, a momentary event, may result in some or multiple missing teeth or supernumerary teeth. A supernumerary tooth is one that is additional to the normal series and can be found in almost any region of the dental arch [2]. Supernumerary teeth may resemble the teeth of the group it may belong, i.e., molars, premolars, or anterior teeth, or it may bear little resemblance in size and shape to the teeth with which it is associated [3]. Teeth in excess of the normal complement are named according to the location: mesiodens are located on the palatal side of the maxillary central incisors; supernumerary canines and premolars are located at the normal sites for such teeth; Para molars located buccal to first, second and third molars and distomolars (fourth molars) located distal to third molars.

The etiology behind supernumerary teeth is not totally understood. It has been suggested that supernumerary teeth develops from a third molar tooth bud arising from the dental lamina near the permanent tooth bud, or possibly from splitting of the permanent bud itself (dichotomy theory) [4]. Another theory is the hyperactivity theory which suggests that supernumerary is formed as a result of local, independent, conditioned hyperactivity of the dental lamina (dental lamina hyperactivity theory). Other theory may describe the presence of supernumerary teeth is the atavism theory (given by Bateson 1894), i.e.; presence of certain ancestral characters in individuals though have become lost in the evolution [5,6] this theory is not accepted currently. Supernumerary teeth may be classified on the basis of its morphology. The four morphological types of the supernumerary are conical (peg shaped), tuberculate (more than one cusp or tubercle often paired), supplemental (duplication of teeth) and odontoma (hamartomatous malformations further divided to compound and complex composite odontoma). The most common supplemental supernumerary is the maxillary lateral incisor, but supplemental premolars and molars may also occur.

This particular condition is found to have increased incidence in cleft lip and palate patients, cleidocranial dysplasia or Gardner's syndrome (multiple impacted supernumerary with polyposis of intestine, multiple sebaceous cyst, and osteomas of bone). A tooth can be stated to be congenitally missing when it is clinically and radiographically missing in the dental arch. Absence of less than six teeth is referred to as hypodontia excluding third molars and absence of more than six teeth is referred to as oligodontia excluding third molars [7,8]. Hypodontia may exist in association with peg shaped laterals, delayed formation and eruption of the other teeth, infra-occlusion of the primary molars, impaction of canines or transposition [9,10], or may be found in isolation. Based on the region of occurrence in the dental arch hypo-hyperdontia can be anterior or posterior thought case with posterior hyper hypodontia have not been reported yet. Previously case of concomitant hypo-hyperdontia was observed [11-14].

Case Report

A 13 years boy came with his father to Department of Orthodontics and dent facial at Govt. College of Dentistry, Indore (M.P., India) with

***Corresponding author:** Bharti C, Assistant professor, Department of Orthodontics, People's College Of Dental Sciences and Research, New Bhanpur Bridge Rd, People's Group, Peoples Campus, Bhanpur, Bhopal, Madhya Pradesh, India, Tel: +91-940-695 5130; E-mail: drchandnibharti@gmail.com

Received January 17, 2018; **Accepted** March 13, 2018; **Published** March 20, 2018

Citation: Bharti C, Ratre RK, Ratre M, Jain S, Singh B (2018) Bi-Maxillary Hyper-Hypodontia: A Unique Case Report with Review of Literature. J Morphol Anat 2: 113.

the chief complaint of inability to close lips and irregular teeth and wanted its correction. The patient's medical history and family history were noncontributory. In occlusion the patient had a mild class II division 2 incisor relationship in the mixed dentition, with an over jet of 2mm and an increased overbite of 4.5 mm (Figure 1). The upper dental midline was shifted to the left by 2 mm with the facial midline while the lower dental midline was coincident.

The lower arch was well aligned; however, both lower permanent central incisors were clinically absent and lower left permanent second molar was erupting. The upper arch was severely crowded with a lack of space for both upper permanent canines. The patient was diagnosed of having supernumerary left central incisors due to which crowding was observed in upper anterior segment and lateral incisor have displaced palatally. Deciduous second molars are present in upper and lower left arch with mobility.

On intra-oral periapical radiographic examination supplemental central incisor is seen to have normal morphological appearance with intact lamina Dura and healthy complete root. Orthopantamo gram shows erupting second molars except second molar in right lower quadrant, which is missing, and no tooth bud is visible. Lower central incisors and all the third molars are also missing. Total number of missing teeth is seven from the full complement of permanent teeth. This is a unique combination of missing teeth and supplemental teeth (Figures 2 and 3).

Treatment

The treatment in the patient was commenced with a full mouth prophylaxis of the patient followed by extraction of the right supplemental central incisor as that was the tooth that leads to elevation of the upper lip leading to the incompetency giving unaesthetic appearance to the patient. The patient was advised to undergo fixed orthodontic treatment for alignment and leveling. Extraction of retained deciduous second molar was performed so that proper eruption pattern can be maintained.

The patient was advised to give follow up after every 6 months to assess the erupting dentition and oral health status. Patient's guardian was explained about the erupting second molars and problems, which may occur due to their supra-eruption, to prevent it later on a RPD, could be given to maintain it. In future, prosthetic implant can be placed in place of second molar.

The patient was given treatment option of undergoing orthodontic treatment after the extraction of the supplemental tooth, along with which replacement of single lateral incisor can be done (Figure 3a).

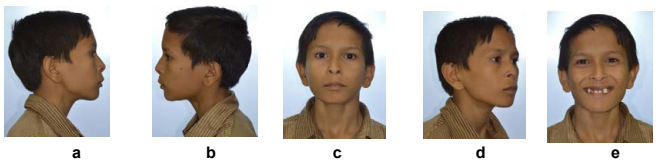

Figure 1: Extra-Oral Photographs.

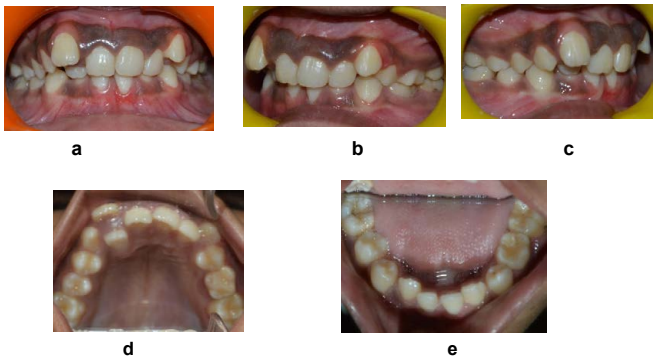

Figure 2: Intra-oral photographs.

Discussion

The exact etiology of hyperdontia is not understood. Some hypothesis has been proposed to explain the formation of supernumerary teeth like atavism, dichotomy, and hyperactivity of the dental lamina and the concept of multi-factorial inheritance. Similarly, some of the theories proposed on the tooth agenesis; like disturbances in differentiation, migration and proliferation of neural crest cells was associated with interactions between the epithelial and the mesenchyme cells during the initiation of ontogenesis may be responsible for hypo-hyperdontia. Few cases of bilateral supplemental maxillary central incisors were described in the literature [15-21]. Gibson reported agenesis of third molars along with a supernumerary tooth in maxilla (mesiodens). Missing mandibular permanent centrals with the presence of supernumerary teeth in maxilla were also reported [22,23]. Hypodontia of mandibular permanent centrals with the presence of midline supernumerary tooth in mandible was reported by many authors [24-27].

Supernumerary most commonly found in the maxillary arch is mesiodens followed by mandibular supernumerary premolars. Supplemental incisors are less common and among incisors supplemental maxillary lateral incisors seem to be more commonly found than central incisors when literature is reviewed (Figure 3b). Mandibular second premolars are the most commonly missing teeth after third molars. Hypodontia found in isolation or associated with some other dental anomaly commonly affects the third molars (9-30%) Followed by maxillary lateral incisors, mandibular incisors and mandibular second premolar. Prevalence of hypodontia in permanent dentition may vary from 2.2 -10.1 % excluding the third molars [28-30] while prevalence of 4.2 % was noted by Gupta et al. [31].

Many cases are reported on concomitant hypo-hyperdontia (CHH). A literature review on the hypohyperdontia shows prevalence ranges from 0.002% to 3.1% one epidemiological study on orthodontic population finds incidence of 0.3% in Non-syndrome CHH [32]. Supernumerary are more common in males while hypodontia is more common in females, CHH has been reported to affect both genders equally [33]. The hypodontia and hyperdontia represent opposite ends of the developmental scale of the dentition showing a gender pre-dominance in males (58 %) than in females, with a 1.3:1 ratio

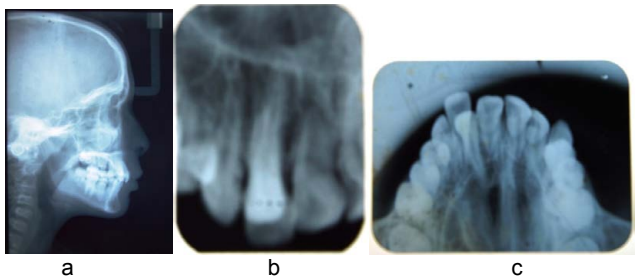

Figure 3: Lateral Cephalogram, Maxillary Occlusal View, Iopa. (a) Lateral Cephalogram (b) Maxillary Occlusal View (c) Intra oral Peri-Apical View.

[34]. Gibson classified this condition as bi-maxillary, maxillary, pre-maxillary (right lateral incisor to left lateral incisor) and mandibular. The syndromes found in association with hyper hypodontia are Down syndrome, Dubowitz syndrome, Ellis-van Creveld syndrome, fucosidosis, G/BBB syndrome, Marfan syndrome, bilateral cleft lip and palate, and cleft palate and abnormalities of the cervical vertebrae [35-38].

Basdra and Magdalini [38] conducted a study to investigate relationships between different malocclusions such as class III and class II division 1 and congenital anomalies. Results showed no statically difference in the occurrence rates of upper lateral incisor agenesis, peg shaped laterals, impacted canines or supernumerary teeth (Figure 3c). When two malocclusions were compared, class III subjects showed significantly higher rates. It can be concluded that there is no genetic relation between congenital anomalies and different malocclusions [39]. Hypodontia is inherited as autosomal dominant mode, but occasionally autosomal recessive and X-linked and polygenic/multifactorial models of inheritance have also been reported. There is large number of genes involved in the odontogenic process, so there are higher chances for mutations to disrupt this process. Tooth agenesis is the most common craniofacial malformation with missing one or more number of teeth (hypodontia, oligodontia or anodontia). Hypodontia can be associated with syndromes or may present as an isolated condition. Hypo-hypertonia is rare in isolation and has been associated with over 50 syndromes (orodigitofacial dysostosis, Hallerman Streiff, Cleidocranial dysplasia syndrome, Ellis van Creveld, Downs's syndrome, cleft lip and palate etc.) [40,41].

Normal development of the tooth germ is appropriately regulated by molecular signaling pathways, if not can give rise to supernumerary/supplemental teeth. These pathways include components of the Hedgehog, FGF, Wnt, TNF and BMP families [42] mutation of genes have been identified by mutational analysis as the major causes of non-syndrome hypodontia (PAX9, MSX1 and AXIN2) [43]. Hypodontia is resulted from haplo-insufficiency of anyone of these and point mutations can cause some or multiple missing teeth. Expression of transcription factors and signaling molecules operating both intracellular and extracellular are guiding tooth development throughout the odontogenic process. BMP4 (transforming growth factor-beta family) and the transcription factors PAX9 (paired box domain) and MSX1 (home box domain), are examples of controlling factors during the odontogenic process. Expression of PAX9 has been found at the sights of tooth development prior to their being any morphological signs of ontogenesis [44].

Mandibular central and lateral incisors show lowest incidence of permanent teeth agenesis, also permanent teeth agenesis of maxillary central incisors, maxillary cuspids and maxillary first molars also rare [45].

Different phenotypic forms in tooth development are the result of different genes involving different interacting molecular pathways, providing an explanation not only for the wide variety of agenesis patterns and also for associations of dental agenesis with other oral anomalies. Different genes involved in human non-syndrome hypodontia includes genes encoding a signaling molecule (TGFA) and transcription factors (MSX1 and PAX9) that are critical during early craniofacial development, but also genes coding for a protein involved in canonical Wnt signaling (AXIN2), and a trans-membrane receptor of fibroblast growth factors (FGFR1) [46]. In a study, frame shift mutation of Pax9 gene (chromosome 14) was identified as responsible for autosomal dominant oligodontia in a large family for four generations. In some of the affected members, maxillary and mandibular second premolars and mandibular central incisors were absent in addition to the lack of permanent molars; although, a normal primary dentition was present [47,48]. Different mutations in the same gene can result in hypodontia or oligodontia, hens these conditions are not fundamentally different. Further, there are evidence to show that more severe the hypodontia, the smaller the Mesiodistal width of the teeth formed, conversely, patients with supernumerary teeth tend to have significantly larger maxillary central and lateral incisors. The size of teeth in case of hypo-hyperdontia is not documented in the literature (Figure 4).

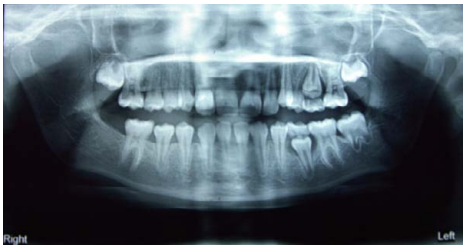

Figure 4: Orthopantomogram.

Conclusion

It is a unique case of supplemental/supernumerary maxillary central incisor and missing mandibular central incisors along with missing right lower second molar and all third molars. This type of case is not reported in the literature which shows counter teeth are affected. In these cases a wise treatment plan is necessary to alleviate patient's problems.

References

1. Verma KG, Vera P, Singh N, Bansal R, Khosa R, et al. (2013) Non-syndromic hypo-hyperdontia - a rare case report and review of literature. Open Journal of Stomatology 3: 37-41.
2. Wise GE, Frazier-Bowers S, D'Souza RN (2002) Cellular, molecular, and genetic determinants of tooth eruption. Crit Rev Oral Biol Med 13: 323-334.
3. Rajendran A, Sivapathasundharam B (2009)Shafer's Textbook of oral pathology, p: 45.
4. Primosch RE (1981) Anterior supernumerary teeth assessment and surgical intervention in children. Pediatr Dent 3: 204-215.
5. Miles AEW, Caroline G, Colyer JF(1936) Variations and Diseases of the Teeth of Animals, p: 6.
6. Polder BJ, Vant Hof MA, Van der Linden FP, Kuijpers Jagtman AM (2004) a meta-analysis of the prevalence of the prevalence of the dental agenesis of permanent teeth. Community Dent Oral Epideol 32: 217-226.
7. Nunn JH, Carter NE, Gillgrass TJ, Hobson RS, Jepson NJ, et al. (2003) The interdisciplinary management of hypodontia: background and role of pediatric Dentistry. Br Dent J 194: 245-251.
8. Arte S, Pirinen S (2003) Hypodontia.Orphanet.
9. Goya HA, Tanaka S, Maeda T, Akimoto Y (2008) An orthopantomographic study of the hypodontia in permanent teeth of Japanese pediatric patients. J Oral Sci 50: 143-150.
10. Gomes RR, da Fonseca JA, Paula LM, Faber J,Acevedo AC (2010) Prevalence of hypodontia in orthodontic patients in Brasilia, Brazil. Eur J Orthod 32: 302-306.
11. Low T (1977) Hypodontia and supernumerary tooth: Report of a case and its management. Br J Orthod 4: 187-190.
12. Gibson AC (1979) Concomitant hypo-hyperodontia. Br J Orthod 6: 101-105.
13. Das G, Sarkar S, Bhattacharya B, Saha N (2006) Coexistent partial anodontia and supernumerary tooth in the mandibular arch: A rare case. J Indian Soc Pedod Prev Dent 24 1: S33-S34.

14. Nuvvula S, Kiranmayi M, Shilpa G, Nirmala S (2010) Hypohyperdontia: Agenesis of three third molars and mandibular centrals associated with midline supernumerary tooth in mandible. Contemp Clin Dent 1: 136-141.

15. Nuvvula S, Pavuluri C, Mohapatra A, Nirmala SVSG (2011) Atypical presentation of bilateral supplemental maxillary central incisors with unusual talon cusp. 29: 149-154.

16. Camilleri S (2003) A case of bilateral supplemental maxillary central incisors. Int J Paediatr Dent 13: 57-61.

17. Rock WP (1991) a case of bilateral supplemental maxillary central incisors. Int J Paediatr Dent 1: 155-158.

18. Stafne EC (1931) Supernumerary upper central incisors. Dental Cosmos 73: 976-980.

19. Steelman R, Wilson C, Nelson S (1991) Maxillary incisor duplication. Oral Surg Oral Med Oral Pathol 71: 523.

20. Tinn CA (1940) Excess, deficiency, and gemination in the deciduous and permanent dentitions of school children. Br Dent J 68: 236-238.

21. Trotman CA, McNamara T (1994) four maxillary incisors: A case report. Spec Care Dentist 14: 112-115.

22. Symons AL (1992) Ectopic eruption of a maxillary canine following trauma. Endod Dent Traumatol 8: 255-258.

23. Anthonappa RP, Lee CK, Yiu CK, King NM (2008) Hypohyperdontia: Literature review and report of seven cases. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 106: 24-30.

24. Low T (1977) Hypodontia and supernumerary tooth: Report of a case and its management. Br J Orthod 4: 187-190.

25. Karthik V, Muralikrishnan B, Anantharaj A (2011) Mandibular mesiodens with agenesis of central incissors (Hypohyperdontia): A Case Report & Review. International Journal of Contemporary Dentistry 2: 26-30.

26. Nayak AG, Chhaparwal Y, Pai KM, Lele AS (2010) Non-syndromic hypo-hyperdontia of the permanent dentition with involvement of the mandibular anterior region: A rare occurrence. Revista de Clínica e Pesquisa Odontológica 6: 281-284.

27. Marya CM, Sharma G, Parashar VP, Dahiya V, Gupta A (2012) Mandibular midline supernumerary tooth associated with agenesis of permanent central incisors: A diagnostic conundrum. Stomatologija 14: 65-68.

28. Mattheews N, Dermaut L, Martens G (2004) Has hypodontia increased in Caucasians during the 20th century? A meta-analysis. Eur J Orthod 26: 99-103.

29. Chung CJ, Han JH, Kim KH (2008) The pattern and prevalence of hypodontia in Koreans. Oral Dis 14: 620-625.

30. Ng'ang'a RN, Ng'ang'a PM (2001) Hypodontia of permanent teeth in a Kneyan population. East Afr Med J 78: 200-203.

31. Gupta SK, Saxena P, Jain S, Jain D (2011) Prevalence and distribution of selected developmental dental anomalies in an Indian population. J Oral Sci 53: 231-238.

32. Varela M, Arrieta P, Ventureira C (2009) Non-syndromic concomitant hypodontia and supernumerary teeth in an orthodontic population. Eur J Orthod 31: 632-637.

33. Mallinemi SK, Nuvvula SK, Cheung CH, Kunduru R (2014) A comprehensive review of the literature and data analysis on the hypo-hyperdontia. 56: 295-302.

34. Chow KM, O'Donnell D (1997) Concomitant occurrence of hypodontia and supernumerary teeth in patient with Down syndrome. Spec Care Dentist 17: 54-57.

35. Hattab FN, Yassin OM, Sasa IS (1998) Oral manifestations of Ellis van Creveld syndrome: report of two siblings with unusual dental anomalies. J Clin Pediatr Dent 22: 159-165.

36. Acerbi AG, de Freitas C, de Magalhaes MH (2001) Prevalence of numeric anomalies in the permanent dentition of patients with Down syndrome. Spec Care Dentist 21: 75-78.

37. Mallinemi SK (2014) Supernumerary teeth: review of literature with recent updates. Conference papers in science.

38. Basdra K, Magdalini NK, Komposch G (2001) congenital tooth anomalies and malocclusions: a genetic link? Eur J Orthod 23: 145-151.

39. Low T (1977) Hypodontia and supernumerary tooth: Report of a case and its management. Br J Orthod 4: 187-190.

40. Pemberton TJ, Das P, Patel PI (2005) Hypodontia: genetics and future perspectives. Braz J Oral Sci 24: 695-706.

41. Zhu JF, Crevoisier R, Henry RJ (1996) congenitally missing permanent lateral incisors in conjunction with a supernumerary tooth: Case report. Pediatr Dent 18: 64-66.

42. Fleming PS, Xavier GM, DiBiase AT, Cobourne MT (2010) Revisiting the supernumerary: the epidemiological and molecular basis of extra teeth. Br Dent J 208: 25-30.

43. Matalova E, Fleischmannova J, Sharpe PT, Tucker AS (2008) Tooth agenesis: from molecular genetics to molecular dentistry. J Dent Res 87: 617-623.

44. Neubuser A, Peters H, Balling R, Martin GR (1997) Antagonistic interactions between FGF and BMP signaling pathways: a mechanism for positioning the sites of tooth formation. Cell 90: 247-255.

45. Dermaut LR, Goeffers KR, De Smit AA (1986) Prevalence of tooth agenesis correlated with jaw relationship and dental crowding. J Orthod Dentofacial Orthop 90: 204-210.

46. De Coster PJ, Marks LA, Martens LC, Huysseune A (2009) Dental agenesis: genetic and clinical perspectives. J Oral Pathol Med 38: 1-17.

47. Stockton DW, Das P, Goldenberg M, D'Souza RN, Patel PI (2000) Mutation of PAX9 is associated with oligodontia. Nat Genet 24: 18-19.

48. Brook AH, Elcock C, Al-Sharood MH, McKeown HF, Khalaf K, et al. (2002) further studies of a model for the etiology of anomalies of tooth number and size in humans. Connect Tissue Res 43: 289-295.