MANDIBULAR HYPO-HYPERDONTIA: A RARE CASE REPORT AND ITS MANAGEMENT

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Abstract
Dental anomalies of tooth number influencing the development of permanent dentition are quite common, and more numerous than in the case of primary dentition, however, combined occurrence of hypodontia and hyperdontia is a rare phenomenon, especially in the same dental arch. The purpose of the present report is to describe a case of concomitant hypo-hyperdontia (CHH) in an 8 year-old male with missing mandibular central incisor and an erupted mandibular mesiodens, and its minimally invasive management. This is a unique case of CHH in the mandible in the dental literature, the two anomalies being manifested in the anterior region of the mandible. Furthermore, this is the only case report describing the evidence of taurodontism in association with mandibular CHH.

Keywords: hypodontia, hyperdontia, mandible, taurodontism

1. INTRODUCTION
Agenesis of one or more teeth in primary or permanent dentition is known as hypodontia, whereas hyperdontia is a condition of having extra teeth. Hypodontia and hyperdontia are two extremes in the development of dentition.

Concomitant occurrence of both hypodontia and hyperdontia (hypo-hyperdontia) in the same patient involves mixed numerical variation. Although the existing literature shows exclusively either hypodontia or hyperdontia, however, only a few surveys have described the occurrence of these numeric anomalies in the same person [1-3]. The term “concomitant hypo-hyperdontia” (CHH) has been used to describe hypodontia and hyperdontia in the same individual [4].

The reported prevalence ranges from 0.002% to 3.1%, most of the reported cases of CHH - identified either accidentally and/or by routine examination - involving both the maxillary and the mandibular arches [1]. All existing reports on CHH refer either to a single arch or to both arches, the maximum number of cases occurring in the maxillary arch. The presence of CHH in the same dental arch in a healthy patient has been reported as a rare phenomenon, extremely rare in the mandibular arch [5]. The present study describes a CHH case in the mandible, and its management with minimally invasive dentistry.

2. CASE REPORT
An 8 year-old boy attended the Department of paedodontics and preventive dentistry for regular dental check-up. His medical, family and personal history was common, revealing that teeth 73 and 83 were extracted due to tooth decay. Extra-orally, he presented a symmetrical face and competent lips. At the time of his presentation in the dental office, he had mixed dentition, and his oral hygiene was inadequate – as he used to brush his teeth once a day. Intra-oral examination showed no abnormality of the soft tissues, except for a generalized gingival inflammation. In the mandibular arch, a microdontic and conical-shaped tooth was noticed in tooth 41 region (Figure 1). Panoramic radiographic examination (Figure 2) confirmed the missing left mandibular central incisor and both maxillary third molars, while a periapical radiography (Figure 3) of the mandibular anterior region showed a close apposition of mesiodens with incisor’ roots. The mesiodens exhibited complete root formation with no evidence of any pathologic periapical changes. Taurodontism was evident in all permanent first molars.
Radiographic examination confirmed the clinical findings and revealed the presence of a normally-oriented erupted mesiodens in relation to tooth 41.

The patient and parents were informed and the treatment options - including: no treatment, composite build-up for supernumerary tooth, extraction of mesiodens and Maryland bridge in relation to tooth 41, no intervention on the tooth until patient’s growth is over, and subsequent implants - were explained to them. The patient preferred a composite build-up of the mesiodens (Figure 4). The child was regularly observed for further multidisciplinary treatment planning.

3. DISCUSSION

The simultaneous occurrence of CHH in the mandible is an extremely rare anomaly in human dentition. The exact etiology of this condition is unknown. The role played by certain specific genes or enzyme defects has not yet been ascertained [1-3]. The etiology of CHH remains unknown and it is not clearly stated whether a specific gene or an enzyme defect might be responsible for the development of this extremely rare condition. Genetic [6] and environmental factors [1] have been postulated to explain this anomaly. Furthermore, CHH has been reported in patients with Down syndrome, Dubowitz syndrome, Ellis–van Creveld syndrome, fucosidosis, G/BBB syndrome, Marfan syndrome and other conditions, such as cleft lip and palate [3].

Based on the site of its occurrence, Gibson [7] classified hypo-hyperdontia as pre-maxillary,
maxillary, mandibular, and bimaxillary. The occurrence of CHH is very unusual in the same arch and extremely rare in the same area of the arch, while mandibular hypo-hyperdontia is relatively rare. Most recently, Zadurska and co-workers [5] reported the occurrence of CHH in both jaws or in the maxilla, but never only in the mandible, concluding that the genetic pattern remains unclear. However, up to now, only few cases have been reported on CHH manifested in the anterior region of the mandible [2,8-13]. In all these cases, mandibular two incisors were missing, and the presence of mesiodens was evident in the mandibular arch, excluding third molars. On the contrary, in the present case, tooth 41 was missing and the supernumerary one was a midline mandibular tooth, a rare occurrence by itself [14-15]. The presence of a microdontic and conical-shaped tooth was evident. Based on its features, the microdontic conical-shaped tooth was diagnosed as a supernumerary tooth. Moreover, in the present case, only one incisor was missing, while the presence of a supernumerary tooth in this position may be concurrent and considered as a novel finding.

In Asian populations, mandibular incisors are the most commonly missing teeth, followed by the mandibular second premolars [16]. The here presented case also had one missing mandibular incisor; however, the patient exhibited developmental absence of two maxillary third molars. If considering the third molars, the case could be viewed as bi-maxillary CHH. However, the congenital absence of the third molars tooth germs in the maxillary arch has not been considered [17]. Hence, this could be an unique example for mandibular CHH. Usually, patients with CHH present no symptoms and are usually detected during examination of other causes or radiographically. When any numeric anomaly of dentition is observed, thorough clinical intra-oral examinations and radiographic investigations are recommended.

Taurodontism has been frequently reported, its incidence rate ranging from 0.25% to 18% [18]. The associations among taurodontism and hypodontia have been reported by several authors. Seow and Lai [19] stated that 34.8% of the patients affected by hypodontia had taurodontism, while a Brazilian study mentioned an one third value [20]. Most recently, the association among CHH and taurodontism has been documented [21]; similarly, in the present case, all four first permanent molars exhibited taurodontism, which might be a coincidental finding, as recently reported. Also, dense vaginatus [22] and double tooth [23] have been reported in association with CHH.

CHH management is quite challenging, since the standard treatment protocols have not been discussed in literature and a multidisciplinary approach is necessary. In the present case, the treatment options were: no treatment, extraction of mesiodens and close space with fixed orthodontics, extraction of mesiodens and Maryland bridge as short-term and implants in feature, and composite build-up to mimic the incisor. Early diagnosis is a key for a successful management, since it permits the dentist to implement the most appropriate treatment options for the patient to minimize consequences. In the present case, composite build-up was done for mesiodens for aesthetic purposes, as preferred by both patient and parents. The treatments may differ from individual to individual, based on the clinical situation. Panoramic radiographs are essential for the diagnosis of hypodontia and/or supernumerary teeth.

4. CONCLUSIONS

CHH is a rare condition, which might be diagnosed by regular clinical and radiographic examination. Agenesis of unilateral mandibular incisor with the presence of mesiodens is very rare in the same individual. Our report highlights a rare occurrence of CHH in an unusual site, and its simple and original management. The presence of CHH with agenesis of single mandibular incisor and presence of mesiodens represents an unique situation, a most rare occurrence. Furthermore, this is the only case report describing taurodontism in association with mandibular CHH.

References

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