Management of unilateral non-syndromic concomitant hypodontia and supernumerary teeth in the mandibular molar region: a rare case report

Susmita Ghosh[1], Paras Mull Gehlot[1], Usha Hegde[2]

[1] Department of Conservative Dentistry and Endodontics, JSS Dental College and Hospital, JSS Academy of Higher Education and Research, Sri Shivarathreeshwara Nagar, Mysuru-570015, Karnataka, India

[2] Department of Oral Pathology, JSS Dental College and Hospital, JSS Academy of Higher Education and Research, Sri Shivarathreeshwara Nagar, Mysuru- 570015, Karnataka, India

SUMMARY

Concomitant hypo-hyperodontia (CHH) is a rare condition that presents with the simultaneous occurrence of hypodontia and supernumerary teeth in the same individual. Even though the exact cause of its occurrence is not known, the combination of two opposite developmental disorders appearing together is mainly attributable to a genetic and environmental cause. In the general population, CHH is rare, and very few cases are published in the literature. This case report hereby presents an incidental finding of a missing mandibular first molar and an atypical unilateral supernumerary parapremolar in a 30-year-old male patient.

Key words: Hypodontia – Supernumerary teeth – Homeobox gene

INTRODUCTION

Disruptions in the initial phases of tooth development can lead to the absence of one or multiple teeth, either through developmental issues or congenital factors. This condition is known as hypodontia, a term commonly used to encompass the entire range of the disorder, ranging from the absence of a single tooth to the exceedingly rare absence of all teeth (Hobkirk et al., 2011).

With the exception of third molars, 5% of populations have at least one missing tooth, ranging from 2.2% to 7.7% (Polder et al., 2004).

The literature contains a wide variety of reports regarding the prevalence of hypodontia, with estimates ranging from 2.6 to 11.3% for the permanent dentition (Larmour et al., 2005). Females are more likely than males to have hypodontia of the permanent dentition, with a ratio of roughly 3:2 (Polder et al., 2004). The least likely teeth to be missing and their prevalence are the maxillary

Corresponding author:
Paras Mull Gehlot, MDS, DIBE, Reader. Department of Conservative Dentistry and Endodontics, JSS Dental College and Hospital, JSS Academy of Higher Education and Research, Sri Shivarathreeshwara Nagar, Mysuru-570015, Karnataka, India. Phone: +919845854977. E-mail: dr.parasmullj@jssuni.edu.in

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central incisor (0.005%), the mandibular canine (0.02%), and the first mandibular molar (0.01%) (Hobkirk et al., 2011; Polder et al., 2004). Hypodontia may also be linked to syndromes or other dental anomalies, including cleft lip and palate (Larmour et al., 2005).

Supernumerary teeth may be defined as any teeth or tooth in excess of the usual configuration of deciduous and permanent teeth. Such a surplus can also be accompanied by a deficit of other teeth (Schulze, 1970). They may occur in both dentitions, unilaterally or bilaterally, singly or multiply (Scheiner and Sampson, 1997). According to orientation, supernumerary teeth can be vertical, inverted, or transverse (Polder et al., 2004). The presence of these teeth may result in various clinical problems, like failure of eruption of adjacent teeth, displacement, crowding, and dentigerous cyst formation (Larmour et al., 2005). The prevalence of supernumerary teeth is approximately 2.1% in permanent dentition (Larmour et al., 2005).

In contrast to other supernumeraries, parapremolar supernumerary teeth that are present in the premolar region tend to occur more frequently in the mandible and are typically of the supplemental type. They can occasionally be smaller than typical premolars or conical in shape. The prevalence of these teeth is reported to be between 0.01 and 1%, depending on the population studied (Khalaf et al., 2018).

The aetiology of supernumerary teeth has been the subject of several theories; however, the evidence points to a multifactorial inheritance pattern that causes the oral lamina to become hyperactive (Rajab and Hamdan, 2002). Moreover, it has been proposed that supernumerary premolar teeth originate from extensions of the dental lamina and form a third (post-permanent) series (Khalaf et al., 2018). In terms of permanent dentition, males are affected around twice as much as females (Schulze, 1970).

The simultaneous occurrence of both conditions, hypodontia and supernumerary teeth, in the same individual is termed 'concomitant hypo-hyperodontia' (CHH) or oligopleiodontia (Varela et al., 2009). There is little published research on the prevalence of CHH in the general population. This paper reports a very rare case of concomitant hypo-hyperodontia, occurring unilaterally in the mandibular arch.

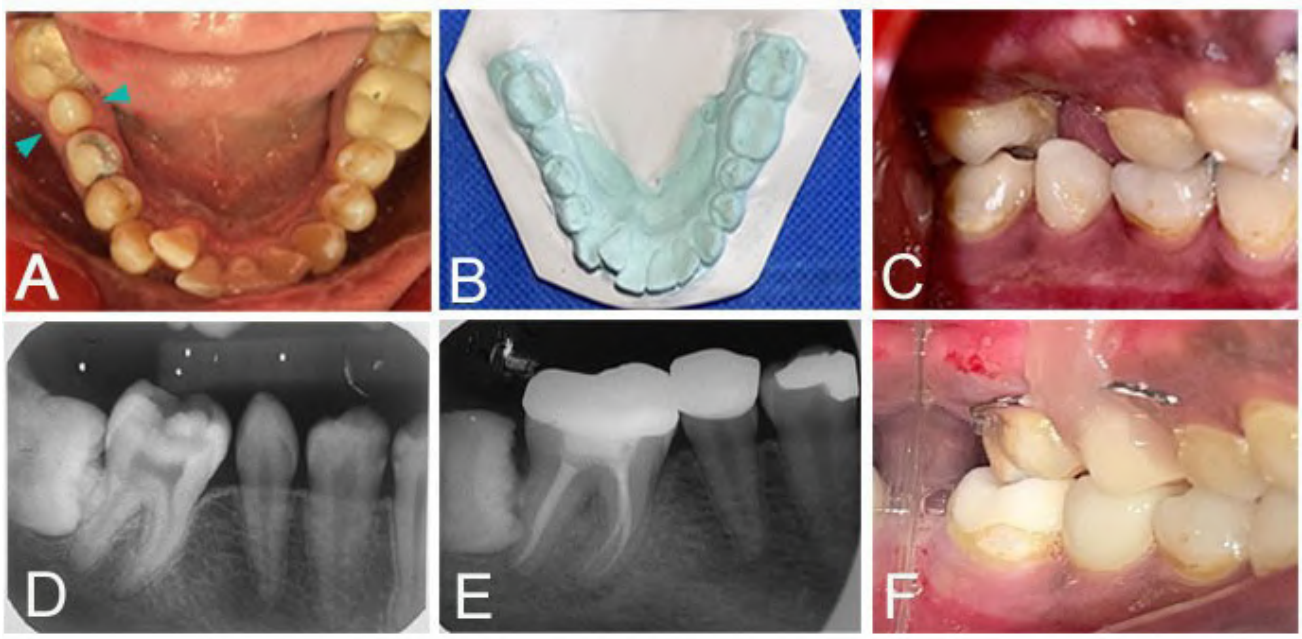

Fig. 1.- A: Intra-oral view of #47 with adjacent missing first molar (#46) and presence of supernumerary Parapremolar (Green arrow heads). **B:** Cast model of the mandibular arch. **C:** Buccal view of the supernumerary tooth. **D:** Pre-operative Intraoral periapical radiograph. **E:** Post-operative Intraoral periapical radiograph. **F:** Postoperative buccal view after post-endodontic restoration.

CASE REPORT

A 30-year-old male reported to the department of conservative dentistry and endodontics with a chief complaint of pain and food lodgement in the lower right back tooth region for 1 month. Past dental history revealed restoration in the lower right molar tooth; however, no history of extraction was reported in the same jaw, although patients reported a history of extraction in the upper molar region. A clinical examination revealed a temporary restoration in tooth no. 47 (Fig. 1A) The tooth was tender on percussion. An intraoral periapical radiograph revealed caries involving pulp with periapical changes (Fig. 1D). Other findings clinically and radiographically revealed a missing mandibular first molar, which was substituted by a supernumerary parapremolar (Fig. 1 A-D).

An OPG (Orthopantomogram) confirmed the clinical findings and the unilateral presentation of CHH (Fig. 2) There was no radiographic evidence of impacted #46 and no history of extraction, which confirmed the congenital absence of #46. The patient was informed about the presence of a supernumerary tooth and the reasons for food lodgement. Other radiographic findings include missing #16 (which was extracted due to its carious destruction), bridge in upper anterior teeth, endodontically treated #27 and #36, and impacted lower third molars and missing # 18.

Informed consent was obtained for non-surgical root canal treatment of #47, followed by a metal-free on lay and a crown on the parapremolar to restore function and occlusion (Fig. 1E, F). As a provisional treatment, the missing #16 was rehabilitated with an acrylic removable partial denture until the patient decided on a definite treatment of an implant or a bridge (Fig. 1F).

DISCUSSION

The etiology of CHH, a combination of two conditions that can be considered opposite developmental disorders, is unknown (Ranta, 1988). The central incisors, canines, first premolars, and first molars are among the “key teeth,” or pole teeth, that are the most mesial teeth in each morphological series, according to Butler’s Field Theory for Evolutionary Development as put forward by Dahlberg (Butler, 1939; Dahlberg, 1945). Since they are thought to be the most genetically stable, they are rarely absent. There was an observation that there was more phenotypic variance in the teeth positioned distally than mesially, as they typically mature later than teeth positioned mesially.

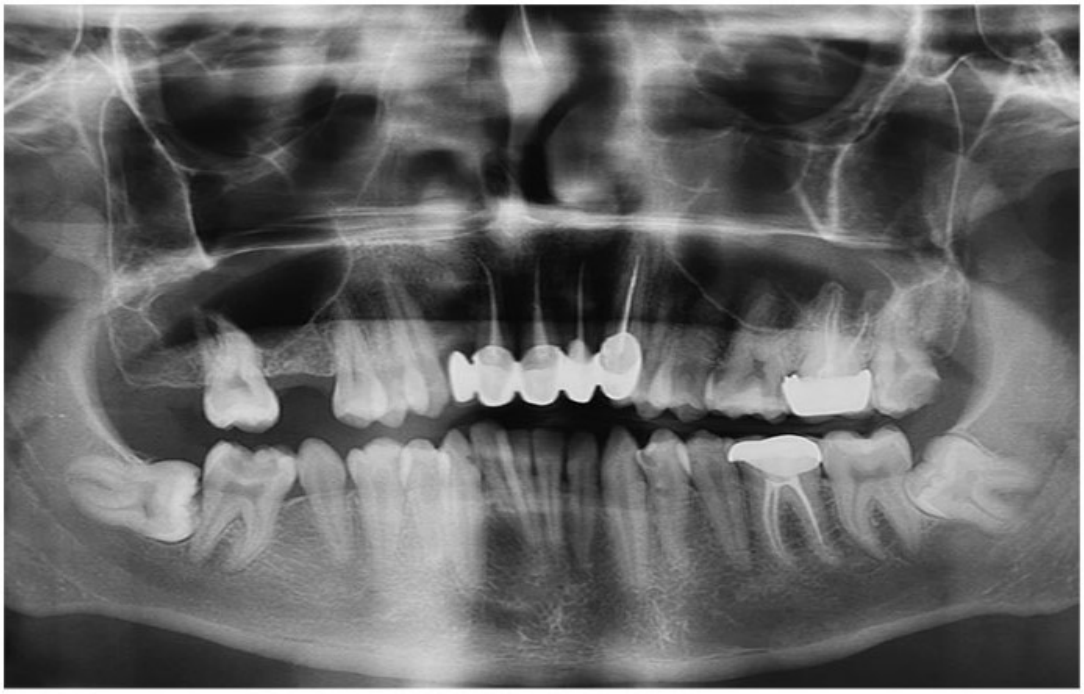

Fig. 2.- Oral pantomogram.

In the present case, the parapremolar was placed distal to the second premolar. Townsend et al. considered a multifactorial model with genetic, epigenetic, and environmental influences, which provides the best explanation for observations involving hypodontia and supernumerary teeth (Townsend et al., 2009; Thesleff, 2000).

It could be caused by interactions between the mesenchymal and epithelial cells during the beginning of odontogenesis, or it could be the consequence of disruptions in the migration, proliferation, and differentiation of neural crest cells (Ranta, 1988). Furthermore, studies using experimental methods have shown that, by modifying specific signalling molecules, it is possible to change the expression regions of the homeobox gene in the ectomesenchymal tissue, and to change the number, size, shape, and differentiation of teeth (Plikus et al., 2005).

The etiology of hypodontia has been linked to several homeobox genes related to tooth development, such as *Msx1, Pax9*, and *Axin2* (Hobkirk et al., 2011). It has been demonstrated that expression of the signalling protein ectodysplasin (EDA) is significant in defining the size of dental fields, in addition to the significance of homeobox genes in regulating the position of various fields within the growing dentition. (Townsend et al., 2009).

Studies conducted on transgenic mice have indicated that modifications to the intracellular adaptor protein EDAR-binding death domain adaptor (EDARADD), its receptor (EDAR), or EDA may result in variations in the number of teeth. It has been suggested that the size of the molar field in mice rises and leads to the creation of extra teeth if the EDA ligand is overproduced or the EDA receptors are overexpressed (Tucker and Sharpe, 2004). In contrast to over-expression of EDAR and its association with extra teeth, when EDAR signalling is lost, it leads to missing molars (Ohazama et al., 2004).

Regarding the familial aspect of hypodontia, it typically manifests as an isolated diagnosis without a clear family history, indicating that a spontaneous genetic mutation may be the cause (Dhanrajani, 2002). In the present case too, the patient reported no family history of missing teeth. The possibility of atypical microdontia of the first molar was considered, but since the morphological appearance of either the crown or root was not in favour of this, the most likely diagnosis was that of supernumerary parapremolar.

While congenital missing teeth (CMT) is not usually considered a serious health issue, hypodontia can have negative effects such as difficulty in articulating speech, poor appearance, periodontal damage, and insufficient growth of the jaw bone (Arif et al., 2024)

The eruption and alignment of the normal dentition may be hampered by the supernumerary teeth. After they are identified, it must be decided whether to extract or leave the additional premolar teeth in place. If a supernumerary premolar is erupted in a reasonable alignment and with no major consequences for the occlusion, then it can be left in place, as in the present case.

It is the clinician's duty to completely inform the patient and/or guardian of all relevant and practical treatment alternatives, along with the advantages and disadvantages of each, as part of the consent procedure.

CONCLUSION

The simultaneous occurrence of hypodontia involving the missing mandibular first molar and the presence of parapremolars (supernumerary) in the first molar space on the same side is unusual and rarely reported. The non-syndromic occurrence in the present case was managed by prosthetic rehabilitation. Early diagnosis and necessary investigations could minimize the risks of complications.

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