CASE REPORT

Bimaxillary concomitant hypohyperdontia in a 10-year-old child

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SUMMARY

Numerical anomalies, either addition or deletion, are quite a common findings in human dentition. However, it is extremely rare to find both hypodontia and hyperdontia simultaneously in the same individual. This condition is referred as concomitant hypohyperdontia (CHH). Aetiology of this condition is still obscure. The prevalence of CHH has been reported to be between 0.002% and 3.1%. This case report highlights a rare occurrence of bimaxillary CHH represented by the absence of both mandibular central incisors and presence of two supernumerary teeth in the maxillary anterior segment. The rarity of such condition of mixed hypodontia as well as hyperdontia in single human dentition prompted the author to report the case.

BACKGROUND

Numerical anomalies, either addition or deletion, are quite a common finding in the human dentition.[1] Hypodontia is a condition when one or few teeth are missing than the normal set of teeth, whereas hyperdontia refers to the presence of an excess number of teeth than the normal set.

Concomitant partial hypohyperdontia (CHH) is a rare condition of human dentition representing mixed numerical anomalies. The aetiology of this simultaneous hyper-hypodontia is obscure. Alteration of neural crest cell epithelial migration, proliferation, differentiation and intercommunication between the mesenchymal cells and epithelial cells during the tooth development process are some of the proposed aetiological factors for CHH.[2] The rarity of such condition of mixed hypodontia and hyperdontia in single human dentition prompted the author to report the case.

CASE PRESENTATION

A 10-year-old male child reported to the Department of Pedodontics and Preventive Dentistry with a complaint of irregular maxillary and mandibular anterior teeth. There was no history of infection, trauma or any metabolic disorders in his childhood. Medical and family history of the child was not significant. He is a lone child born to parents of a non-consanguineous marriage. No syndromic features were evident in the child on physical examination. The boy had normal built, height and IQ. All vital signs and haematological and serological values were well within the normal limits.

Intraoral examination revealed mixed dentition with adequate oral hygiene. The child brushes his teeth once a day with the horizontal scrub technique. Intraoral examination shows the absence of both the permanent mandibular central incisors (#31, #41) with retained deciduous left central incisor (#71). A conical-shaped tooth present in the midline between maxillary central incisors suggested supernumerary teeth (SNT) (mesiodens) (figure 1), which resulted in a labial shift of maxillary right central incisor and shift in the dental midline. Maxillary left central incisor was in crossbite relation to mandibular left lateral incisor. Occlusal dental caries was present with respect to maxillary left deciduous first molar (#64).

INVESTIGATIONS

On radiographic examination, an intraoral periapical radiograph (IOPAR) of the mandibular anterior region confirmed the agenesis of #31 and #41 with retained #71 (figure 2). The maxillary IOPAR showed the presence of a conical-shaped mesiodens having complete root length and no evidence of any periapical pathology (figure 3). Furthermore another impacted SNT was seen in relation to the roots of maxillary central incisors. An orthopantomogram also confirmed the aforementioned IOPAR findings, that is, the agenesis of #31 and #41 with retained #71 and the presence of two SNT in the maxillary arch, a mesiodens in the midline and an impacted tooth near the roots of maxillary incisors (figure 4).

DIFFERENTIAL DIAGNOSIS

A provisional diagnosis based on the complete history and clinical examination was bimaxillary hypohyperdontia, and the differential diagnoses of orodigitofacial dysostosis, Hallerman-Streiff and cleidocranial dysplasia syndromes were considered. Based on the complete history, clinical examination and radiological investigations, bimaxillary hypohyperdontia was the final diagnosis.

TREATMENT

Based on the findings the treatment advised here was the extraction of two SNT in maxillary arch and retained deciduous mandibular central incisor followed by orthodontic corrections. Although complete treatment plan was explained to the child and the parents, the parents refused to undergo any treatment. The only dental care provided was oral prophylaxis, glass ionomer cement (GIC) restoration with respect to #64 and oral hygiene instructions.

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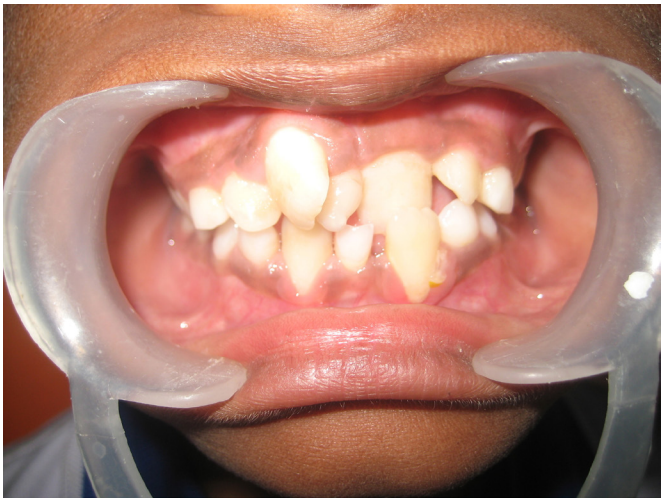

Figure 1 Clinical intraoral image displaying supernumerary tooth in the maxillary arch and missing central incisors in mandibular arch.

OUTCOME AND FOLLOW-UP

On follow-up visit after 1 month, the child had adequate oral hygiene and intact GIC restoration. Parents were once again informed and explained about the importance of treatment, but they still refused to undergo any corrective treatment.

DISCUSSION

CHH is a rare dental condition representing mixed numerical anomalies. CHH can be defined as an extremely rare numerical mixed condition in which the teeth may be supernumerary or absent, relative to the normal complement.[3] The condition is seen in both primary and permanent dentition, and may affect the maxilla and/or mandible. The condition is asymptomatic and usually discovered during the clinical and radiographical examination of the oral cavity for other purposes. The diagnosis of CHH in our case was also an incidental finding. Moreover, it has been stated that most of the patients with CHH may remain undiagnosed.

Many different terminologies have been used in the past to describe this condition, such as 'concomitant hypodontia and hyperdontia CHH'[4] and 'oligopleiodontia'.[5] However, as suggested by Gibson, 'hypohyperdontia' is preferred nowadays.[1]

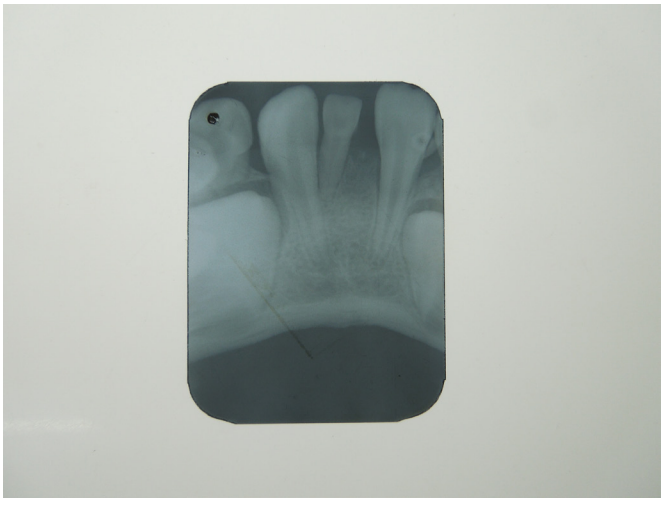

Figure 2 Intraoral periapical radiograph mandibular incisor region confirming missing #31 and #41 along with over-retained #71.

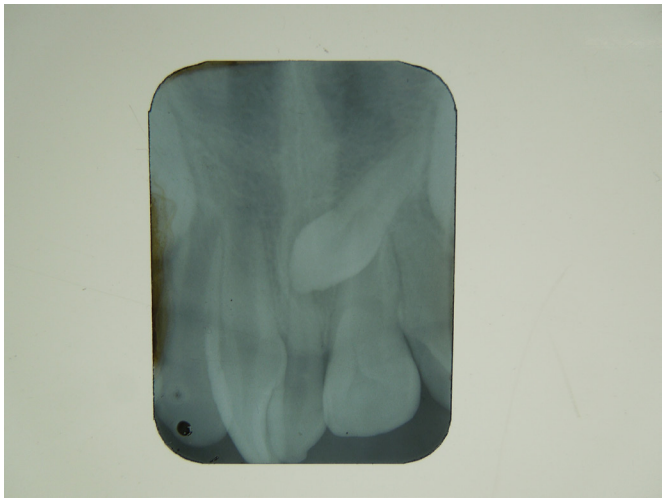

Figure 3 Intraoral periapical radiograph maxillary incisor region confirming presence of two supernumerary teeth.

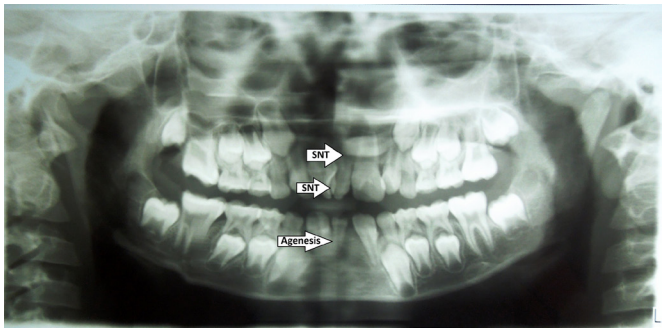

Figure 4 Orthopantomogram showing concomitant hypohyperdontia. SNT, supernumerary tooth.

Classification

Gibson has classified hypohyperdontia into four subdivisions based on the area of involvement, that is, premaxillary, maxillary, mandibular and bimaxillary hypohyperdontia. The present child serves as a good example for bimaxillary hypohyperdontia since hypodontia (agenesis of mandibular central incisors) and hyperdontia (mesiodens and impacted SNT in the premaxillary area) were noted in both the arches,[1] whereas Mallineni *et al*[3] classified it as the maxillary type (the maxillary arch alone), mandibular type (the mandibular arch alone) and bimaxillary type (both the maxillary and mandibular arches).

Prevalence

Hypodontia

The incidence of hypodontia is reported as 3.5%–8% by Polder *et al*[6]; however, the incidence reported in the mandibular central incisor region is 0.41%–0.5% by Low in 1977[7] and 0.2%–0.4% by Robinson *et al*.[8]

Hyperdontia

According to Rajab and Hamdan, the prevalence of SNT in Caucasian population for the permanent dentition ranges from 0.1% to 3.8%.[9] Luten JR in 1967 reported the incidence of maxillary mesiodens to be in the range of 0.15%–3.0%.[10] Anthonappa *et al*[11] reported that the mesiodens is the most common SNT associated with CHH.

Hypohyperdontia

Anthonappa *et al*[11] reported the prevalence of CHH as between 0.002% and 3.1%. Literature suggests male preponderance.[1 3] The most commonly reported CHH is bimaxillary CHH, followed by maxillary CHH, and mandibular CHH being the least prevalent. Reviews have suggested that hyperdontia is more common in the anterior maxillary region and hypodontia is more common in the posterior mandibular region in cases of hypohyperdontia.[3]

Varela *et al* conducted a retrospective study on 2108 non-syndromic orthodontic patients aged 7–16 years and reported CHH prevalence as 0.33%. This study also found a correlation between CHH and some syndromes.[12]

Aetiology

Even though many hypotheses have been speculated for CHH, the exact aetiology still remains obscure.[13] Alteration of neural crest cell epithelial migration, proliferation, differentiation and intercommunication between the mesenchymal cells and epithelial cells during the tooth development process are some of the proposed aetiological factors for CHH.[2] Recently it is suggested that genetic and environmental factors both add to the aetiology of simultaneous occurrences of tooth agenesis and SNT.[14]

Management

The management of CHH is very complex and needs a multidisciplinary approach, as there are no standard treatment protocols documented in the literature. In the present case, the treatment options included extraction of mesiodens and retained deciduous incisor followed by fixed orthodontic correction; however, parents declined for the treatment. The key to successful treatment lies in early diagnosis, as it permits the dentist to enforce the most appropriative treatment at the right time to reduce consequences. Panoramic radiographs are essential for the diagnosis of hypodontia and/or SNT.[3 11]

Syndromes

Hypohyperdontia is frequently associated with various syndromes like bilateral cleft lip and palate, Down syndrome, Ellis-Van Creveld syndrome, cleidocranial dysplasia, Dubowitz syndrome, fucosidosis, Opitz G syndrome, Marfan syndrome and cleft palate and abnormalities of the cervical vertebrae.[3 11] In the present case, the patient was not suffering from any syndrome.

Different reported cases

Camilleri[4] was the first to report two cases of maxillary CHH in 1967. Many authors have reported cases of CHH, such as Sharma, who observed two cases where both patients had premaxillary SNT. One of them exhibited missing mandibular second premolar. The other patient showed agenesis of the permanent mandibular lateral incisor.[15] Agenesis of a mandibular permanent central incisor with mandibular mesiodens was reported by Das *et al*,[16] Gibson[1] and Verma *et al*.[17] Manjunatha *et al*[18] described a case of a 26-year-old man with missing mandibular central incisors and presence of a premaxillary SNT, similar to our case. Anthonappa *et al* also reported a case similar to the present case with missing mandibular permanent centrals with the presence of two SNT in the maxilla.[11]

Learning points

- Concomitant hypohyperdontia is a rare developmental anomaly of number occurring in the human dentition.
- It is diagnosed on routine clinical examination and confirmed by radiographical examination.
- Concomitant hypohyperdontia affects aesthetics and hence patient's confidence and self-esteem.
- Management is multidisciplinary, and may necessitate extractions of extra teeth followed by re-establishing aesthetics and occlusion by orthodontic treatment.

Contributors PBK: Clinical examination of case and history taking, diagnosis, and clinical photographs recording. SS: Radiographic investigation and manuscript writing. NK: Orthodontic consultation and patient counselling, and manuscript editing. AA: Manuscript writing and editing, and review of literature.

Competing interests None declared.

Patient consent Obtained from guardian.

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