Hypohyperdontia: Agenesis of three third molars and mandibular centrals associated with midline supernumerary tooth in mandible

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Abstract

Agenesis of teeth in a patient who also presents with a supernumerary tooth is one of the rare numerical anomalies in human dentition. Agenesis of third molars was shown to be associated with other missing permanent teeth. A review of literature on hypodontia including third molar agenesis, hyperdontia and a concomitant presence of these two conditions which is termed as hypohyperdontia is presented along with a case showing agenesis of three third molars, both mandibular central incisors and a midline supernumerary tooth.

Keywords: Hypodontia, hypohyperdontia, supernumerary tooth, tooth agenesis

Agenesis of teeth in a patient with associated supernumerary tooth/teeth is one of the rare numerical anomalies in human dentition. This numerical anomaly is termed as hypohyperdontia or oligoplieodontia.

Hypodontia

Congenitally missing tooth is defined as the one not erupted in the oral cavity, and also not visible in a radiograph. Hypodontia means that one to six teeth are missing (excluding the third molars). When more than six teeth are missing, the term oligodontia is used, and complete absence of teeth is termed as anodontia. For diagnosis of congenitally missing teeth, the age of examination is 3 – 4 years in primary dentition and 12 – 14 years in permanent dentition.[1] A meta-analysis[2] showed that permanent teeth agenesis differs by continent and gender; the prevalence for both sexes was higher in Europe (males 4.6% to females 6.3%) and Australia (males 5.5% to females 7.6%) than for North American Caucasians (males 3.2% to females 4.6%). In addition, the prevalence of dental agenesis in females is 1.37 times higher than in males. The second mandibular premolar is the most affected tooth, followed by the maxillary lateral incisor, and the second maxillary premolar. Unilateral occurrence of dental agenesis is more common than bilateral occurrence. However, bilateral agenesis of maxillary lateral incisors is more common than unilateral agenesis. The overall prevalence of agenesis in the maxilla is comparable with that in the mandible, but a marked difference was found between both jaws regarding tooth type.

Third molar agenesis was reported by Nanda,[3] Eidelman et al,[4] Thompson et al,[5] and Mathewson et al.[6] In a study on contemporary American adolescents Harris[7] showed that 15% of the sample showed at least one missing third molar and about 2% for all four third molars. Sandhu and Kaur[8] reported a prevalence of 11.5%. Rozkovcová et al,[9] studied the incidence of third molar agenesis in young Czech population and showed an incidence of 22.5% of agenesis of 1 to 4 third molars. The incidence of third molar agenesis was shown to be ranging from 23% to 28%.[9,10] Agenesis of third molars was shown to be associated with absence of lateral incisors (12%), followed by second premolars (11%), central incisors and second permanent molars (3%), and canines and first premolars (1%) in a study population compared to a group with the presence of all four third molars. In this study, the control group having all four third molars showed fewer than 2% missing teeth with only laterals or second premolars involved.[11] It is curious to learn that the occurrence of hypodontia elsewhere in the permanent dentition is 13 times greater than the population prevalence when third molar is congenitally missing.[12]

According to Richardson,[13] third molars are the more commonly missing teeth with a range from 9 to 20% population with one or more third molars missing, and a male to female ratio of 3:2.

There may be several causes for congenital absence of teeth including physical obstruction or disruption of the dental lamina, space limitation, and functional abnormalities of the dental epithelium or failure of initiation of the underlying mesenchyme.[14] Congenital absence of teeth may result from genetic factors as it is described as one of the features in more than 60 syndromes.[15] According to Viera et al,[16] MSX1, TGFA and PAX9 interaction sometimes said to play a role in tooth agenesis.
Hyperdontia

Supernumerary teeth, also termed as hyperdontia, may occur in both the dentitions, unilaterally or bilaterally, and in either or both maxilla and mandible.[17,18]

Literature reports say that the prevalence of supernumerary teeth in permanent dentition ranges from 0.2 to 0.9%. [19] They may be seen in any region of the dental arch with a predilection towards maxilla.[20] In the permanent dentition, the ratio of supernumerary teeth in males is two times more than females.[17] Some Asian surveys show a predominance of male patients[21] with a ratio of 6.5:1.

The crowns of the supernumerary teeth may show either a normal appearance or show different atypical shapes and with mature or immature roots.[22] Two-thirds of supernumerary teeth in primary dentition and one-quarter of those in permanent dentition will erupt normally and the rest may remain unerupted and show some complications.[23,24] There are several theories proposed to explain the etiology of supernumerary teeth, but the available data suggest a pattern of multifactorial inheritance leading to hyperactivity of dental lamina and hyperdontia was seen in over 20 different syndromes.[17]

Hypohyperdontia

Simultaneous occurrence of tooth agenesis (hypodontia) and supernumerary or supplemental teeth (hyperdontia) in the same individual is one of the rare anomalies of number in human dentition. Literature shows that hypohyperdontia may affect the primary and/ or permanent dentition and may involve maxilla and/ or mandible.

The presence of this hypohyperdontia in the same segment of an arch is even rarer occurrence. Various terms were used to denote this condition, coexistence of hypohyperdontia, concomitant hypodontia and hyperdontia,[25] concomitant hypohyperdontia,[26] and oligo-pleiodontia[27] (‘Oligos’ meaning few; ‘Pleion’ meaning more or extra in Greek). In surveys of general population or children attending dental practices,[28,29] the frequency of hypohyperdontia ranged between 8 -15 per 10,000. The prevalence seems to be higher in some Asian (Chinese) populations[21] i.e., 40 per 10,000. Only a few reports in literature[30] have shown the prevalence of associated hypodontia and hyperdontia in the same individuals, which ranged from 0.002% to 3.1%. Males were more commonly reported with this condition even though there seems to be no sexual predilection. Varela et al.[31] in a study of 2108 non-syndromic orthodontic patients reported concomitant hypohyperdontia (CHH) in 7 patients (0.33%) out of which male to female ratio was 4:3. Hypohyperdontia is found more frequently in permanent dentition than in primary and mixed dentitions.[32] There appears to be a correlation between hypohyperdontia and cleft-lip/ palate (CLP) and also some syndromes such as Down,[33,34] Ellis van Creveld,[35,36] and conditions like Fucosidosis[37] which can be explained by the higher prevalence of isolated hypodontia and supernumerary teeth in these conditions.

The etiology of hypohyperdontia is unknown. Several attempts have been made to find the possible interpretation of the association of these two different numerical anomalies. It may result from disturbances in migration, proliferation, and differentiation of neural crest cells during the initiation of odontogenesis.[12]

Gibson[38] reported 20 cases of hypohyperdontia [Table 1] out of which 6 cases were with agenesis of 1 to 4 third molars along with a maxillary mesioden. Literature reports of hypohyperdontia show missing maxillary laterals, mandibular second premolars, and maxillary second premolars more often compared to any other compliment of teeth.[39] The reported cases of associated hypodontia with hyperdontia (supernumerary or supplemental teeth) otherwise termed as hypohyperdontia[25,27,28,30,34,37,39-58] were shown in Table 2.

Case Report

A 15-year-old female patient accompanied by mother, reported to the department of Pedodontics with the chief complaint of decayed tooth in the lower right jaw. This was the first visit to a dentist and the patient gave a history of occasional pain which subsides on taking some medicines from a general physician. There was no significant medical history and the general health appeared to be normal. Intraoral examination revealed permanent dentition (excluding third molars), with presence of mandibular midline supernumerary tooth and absence of permanent mandibular central incisors [Figure 1].

Mandibular right first permanent molar was grossly decayed, with the destruction of lingual wall and the oral hygiene was fair.

An intraoral periapical view [Figure 2] of mandibular
Table 1: Distribution and frequency of teeth involved with concomitant hypo-hyperdontia according to Gibson (1979)[26]

| Case No | Sex | Age (Yrs) | Third molars | Hypodontia | Hyperdontia | Subdivision of concomitance |
|---------|-----|-----------|--------------|------------|-------------|-----------------------------|
|         |     |           | Excluded     | Included   |             |                             |
| 1       | M   | 8         | *            |            | 12          | Maxillary mesiodens         | Premaxillary Maxillary |
| 2       | M   | 9         | *            |            | 22          | Maxillary mesiodens         | Premaxillary Maxillary |
| 3       | M   | 11        | *            |            | 15, 22      | Maxillary mesiodens         | Premaxillary Maxillary |
| 4       | F   | 13        | *            |            | 15, 25      | Maxillary mesiodens         | Maxillary         |
| 5       | F   | 11        | *            |            | 15          | Supplement 22               | Maxillary         |
| 6       | F   | 8         | *            |            | 31, 41      | Mandibular mesiodens        | Mandibular         |
| 7       | F   | 14        | *            |            | 42          | Four Denticles in 45, 46   | Mandibular         |
| 8       | F   | 8         | *            |            | 45          | Supplement 22               | Bimaxillary       |
| 9       | F   | 13        | *            |            | 15, 25, 35  | Maxillary mesiodens         | Bimaxillary       |
| 10      | F   | 10        | *            |            | 45          | Maxillary mesiodens         | Bimaxillary       |
| 11      | M   | 13        | *            |            | 35, 45      | Maxillary mesiodens         | Bimaxillary       |
| 12      | F   | 9         | *            |            | 15, 25, 35, 45 | Supplement 22 | Bimaxillary |
| 13      | M   | 9         | *            |            | 25          | Two supplemental molars in 17 and 26 area | Bimaxillary |
| 14      | M   | 10        | *            |            | 22, 23, 25, 35, 31, 41, 45 | Maxillary mesiodens | Bimaxillary |
| 15      | M   | 14        | *            |            | 48          | Maxillary mesiodens         | Bimaxillary       |
| 16      | M   | 17        | *            |            | 38, 48      | Maxillary mesiodens         | Bimaxillary       |
| 17      | M   | 14        | *            |            | 18, 38, 48  | Maxillary mesiodens         | Bimaxillary       |
| 18      | M   | 15        | *            |            | 18, 28, 38, 48 | Maxillary mesiodens | Bimaxillary |
| 19      | F   | 16        | *            |            | 18, 28, 38, 48 | Maxillary mesiodens | Bimaxillary |
| 20      | F   | 15        | *            |            | 18, 28, 38, 48 | Maxillary mesiodens | Bimaxillary |

central incisors was taken using radiovisiography (RVG) which revealed the absence of both the centrals and presence of a conical supernumerary tooth in the midline with completed root formation. The patient was advised an orthopantomograph (OPG) to check for any other developmental anomalies of teeth and also to evaluate the status of mandibular right first permanent molar. The OPG [Figure 3] revealed agenesis of bilateral maxillary third molars and mandibular right third molar (18, 28 and 48) in addition to mandibular centrals (31 and 41) and a completely developed crown of mandibular left third molar (38). Mandibular right first permanent molar (46) showed inter-radicular bone loss with internal resorption of the pulp chamber. There were no reported cases of any missing teeth in the parents or other family members. The patient was advised oral prophylaxis and extraction of 46, followed by prosthetic rehabilitation of 46. The supernumerary tooth in mandibular midline was planned for esthetic restoration using composite resins.
## Discussion

Simultaneous occurrence of tooth agenesis (hypo/oligodontia) and presence of supernumerary tooth has been reported in the literature. The reports showing associated agenesis of third molars along with a supernumerary tooth in maxilla (mesiodens) are listed in Table 2.

### Table 2: Reported cases of hypohyperdontia*

| Author/s                        | Age   | Gender | Agenesis          | Hyperdontia          | Remarks                        |
|---------------------------------|-------|--------|-------------------|----------------------|--------------------------------|
| Munns[38]                       | 17 yrs| F      | 12, 22            | 43                   | -                              |
| Cammilleri[39]                  | 17 yrs| F      | 12, 22            | Mesiodens            | -                              |
| Brook and Winter[39]            | 10 yrs| F      | 12                | Mesiodens            | -                              |
| Nathanall[37]                   | 11 yrs| F      | 35, 45            | Mesiodens            | -                              |
| Mercer[39]                      | 20 yrs| M      | 15, 35, 45        | Mesiodens            | -                              |
| Low[41]                         | 7 yrs  | M      | 31, 41            | Supernumerary tooth in mandibular midline | -                              |
| Spyropoulos et al.[41](3 cases) |       |        | 15, 14, 12, 22, 24, 25, 35, 37, 44, 45 | Mandibular Incisor | -                              |
|                                 | 15 yrs| M      | 13, 45            | Mesiodens            | -                              |
|                                 | 13 yrs| M      | 31                | Supplemental 22      | -                              |
| Moore[42]                       | 11 yrs| M      | 13, 23            | Supernumerary tooth between 11 and 12 | -                              |
| Ranta[43]                       | 5 yrs  | M      | 53, 63, 74, 34    | Supplemental 72, 32  | -                              |
| Macpherson[37]                  | 6 yrs  | F      | 52, 62            | Supernumerary teeth in 83, 82, 72 region | -                              |
| Symons[44]                      | 8 yrs  | M      | 31, 41            | Supernumerary teeth in 11 and 13 region | -                              |
| Hewson et al.[45]               | 9 yrs  | M      | 45                | Supplemental maxillary central incisor | -                              |
| Zhu et al.[46]                  | 15 yrs| M      | 12, 22            | Supernumerary tooth apical to 46 | -                              |
| Chow and O’Donnell[47]          | 13 yrs| F      | 15, 45            | Supernumerary teeth in 32 and 42 region | Down Syndrome                  |
| Scheiner and Sampson[46]        | 8 yrs  | F      | 35, 45            | 2 mesiodentes        | -                              |
| Segura and Jimenez-Rubero[49]   | 13 yrs| M      | 22                | Mesiodens            | -                              |
| Hattab et al.[50](2 cases)      | 9 yrs  | M      | 12, 22, 31, 32, 33, 42, 43 | Mesiodens | Ellis-van Crevald syndrome  |
|                                 | 8 yrs  | F      | 12, 22, 31, 32, 42, 42 | Supernumerary in 11 region | -                              |
| Sharma[41]                      | 12 yrs| F      | 23                | Supernumeraries in 15, 14, 12, 21, 22, 24, 32, 34, 35, 42 and region | -                              |
| Matsumoto et al.[52]            | 8 yrs  | F      | 25, 32            | Mesiodens            | -                              |
| Acri[41]                        | 12 yrs| M      | 15, 25, 34, 35, 45 | Mesiodens            | -                              |
| Oliveira et al.[33](2 cases)    | 8 yrs  | F      | 35, 45            | 2 mesiodens          | -                              |
| El-bahannasawy and Fung[54]     | 5 yrs  | M      | 53                | Supplemental 54, 15  | -                              |
| Patchett et al.[53](7 Cases)    | 9 yrs  | M      | 35, 45            | Supplemental 11, palatal supernumerary tooth | -                              |
| Das et al.[56]                  | 8 yrs  | F      | 31, 41            | Supernumerary tooth in 31, 41 area | -                              |
| Anthonappa et al.[30]           | 12 yrs| F      | 31, 41            | Supplementary 23     | -                              |
|                                 | 9 yrs  | M      | 15, 35            | Supplementary 21     | -                              |
|                                 | 11 yrs| M      | 32                | Supplementary 15     | -                              |
|                                 | 5 yrs  | M      | 71, 81, 42        | 2 supernumerary teeth in 11, 21 area | -                              |
|                                 | 7 yrs  | M      | 31, 41            | -                    | -                              |
|                                 | 5 yrs  | F      | 72, 32            | Mesiodens            | -                              |
|                                 | 6 yrs  | M      | 35, 45            | Mesiodens            | -                              |
|                                 | 7 yrs  | M      | 25                | 2 mesiodentes        | Identical twins                |
|                                 | 7 yrs  | M      | 35                | Mesiodens            | -                              |
| Lertsirivorakul J[58]           | 9 yrs  | F      | 15, 14, 25, 35, 36, 45, 46, 47 | Supplemental 22 | -                              |

*Cases reported by Gibson (1979) were not included in this Table.
was reported by Gibson.\textsuperscript{[20]} Missing mandibular permanent centrals with the presence of supernumerary teeth in maxilla were reported by Symonds\textsuperscript{[43]} and Anthonappa et al.\textsuperscript{[30]} Hypodontia of mandibular permanent centrals with the presence of midline supernumerary tooth in mandible was reported by Low,\textsuperscript{[40]} Gibson\textsuperscript{[26]} and Das et al.\textsuperscript{[54]}

The association of third molar agenesis with missing teeth from other classes of teeth was shown by Garn et al.\textsuperscript{[11]} According to them the association between third molar agenesis and reduction in the number of other teeth fits the hypothesis of a field of variable intensity, which, in its greatest degree of expression, eliminates all four third molar teeth and a maximum number of other teeth. The association between third molar reduction and developmental delay in the dentition is susceptible to at least two hypotheses, one involving pleiotropic manifestations of a single gene and the other involving two independent genes, the first favoring developmental suppression and the second affecting formation timing. While the degree of independence between these two phenomena may show which hypothesis is correct, the possibility of closely linked genes must also be considered. In this latter event, the monogenic and polygenetic hypotheses would be operationally identical.

According to Poyton et al.\textsuperscript{[93]} the most common type of supernumerary tooth is mesiodens, located between maxillary central incisors followed by maxillary fourth molars, mandibular molars, premolars, cuspids and lateral incisors. The incidence of supernumeraries in the lower incisor region is low and is usually of supplemental type\textsuperscript{[86]} with an occurrence rate of 2\textsuperscript{[6]}.\textsuperscript{[63]} Cases of supernumerary teeth in mandible which are of supplemental type were reported by Sharma\textsuperscript{[62]} and Yokose et al.\textsuperscript{[63]} There was a 56\% incidence of conical-shaped supernumerary teeth with these conical shaped teeth located between the central incisors.\textsuperscript{[64]}

This case presented by us is in accordance with the findings of Garn et al.,\textsuperscript{[11]} and Bailit\textsuperscript{[12]} that third molar agenesis is associated with missing teeth from other classes of teeth. The present case shows agenesis of three third molars (18, 28, and 48) and the mandibular central incisors (31, 41) with the presence of a midline supernumerary tooth (hypohyperodontia), which has never been reported in the literature as per our knowledge [Table 3].

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**Table 3: Details of the presented case**

| Age  | Gender | Agenesis/hypodontia | Hyperodontia | Remarks |
|------|--------|---------------------|--------------|---------|
| 15 yrs | F | 18, 28, 31, 41, 48 | Supernumerary tooth in mandibular midline | - |

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