Non-syndromic occurrence of true generalized microdontia with mandibular mesiodens - a rare case

Seema D Bargale* and Shital DP Kiran

Abstract
Abnormalities in size of teeth and number of teeth are occasionally recorded in clinical cases. True generalized microdontia is rare case in which all the teeth are smaller than normal. Mesiodens is commonly located in maxillary central incisor region and uncommon in the mandible. In the present case a 12 year-old boy was healthy; normal in appearance and the medical history was noncontributory. The patient was examined and found to have permanent teeth that were smaller than those of the average adult teeth. The true generalized microdontia was accompanied by mandibular mesiodens. This is a unique case report of non-syndromic association of mandibular hyperdontia with true generalized microdontia.

Keywords: Generalised microdontia, Hyperdontia, Permanent dentition, Mandibular supernumerary tooth

Introduction
Microdontia is a rare phenomenon. The term microdontia (microdentism, microdontism) is defined as the condition of having abnormally small teeth [1]. According to Boyle, “in general microdontia, the teeth are small, the crowns short, and normal contact areas between the teeth are frequently missing” [2] Shafer, Hine, and Levy [3] divided microdontia into three types: (1) Microdontia involving only a single tooth; (2) relative generalized microdontia due to relatively small teeth in large jaws and (3) true generalized microdontia, in which all the teeth are smaller than normal. According to these authors, aside from its occurrence in some cases of pituitary dwarfism, true generalized microdontia is exceedingly rare. Microdontia of a single tooth can be further classified into (1) microdontia of the whole tooth, (2) microdontia of the crown of the tooth, and (3) microdontia of the root alone [4].

Involvement of the entire dentition is rare and been reported in radiation or chemotherapeutic treatment during the developmental stage of the teeth [5], pituitary dwarfism [3] and Fanconi’s anemia [6]. The syndromes associated with microdontia are Gorlin-Chaudhry-Moss syndrome, Williams’s syndrome, Chromosome d/u, 45X [Ullrich-Turner syndrome], Chromosome 13[trisomy 13], Rothmund-Thomson syndrome, Hallermann-Streiff, Oro-faciodigital syndrome (type 3), Oculo-mandibulo-facial syndrome, Tricho-Rhino-Phalangeal, type1 Branchio-oculo-facial syndrome.

Supernumerary teeth are defined as any supplementary tooth or tooth substance in addition to usual configuration of twenty deciduous and thirty two permanent teeth [7]. Classification of supernumerary teeth may be based on position or morphology. Positional variations include anterior mesiodens, para-premolars, para-molars and distomolars. Variations in morphology consist of supplemental and rudimentary types [8].

Supernumerary teeth are common in the maxillary anterior region although supernumerary teeth have been reported in the incisor region of the mandible are very rare. Although supernumerary teeth have been reported in the incisor region of the mandible, they are very rare [9-14].

Conditions, in which supernumery teeth found, are cleidocranial dysplasia, cleft lip and cleft palate [15]. Syndromes associated with supernumery teeth are Familial adenomatous polyposis [Gardner’s], Apert, Klippel-Trenaunay-Weber, Craniofaciokeletal dysplasia, Trisomy 21 [Down’s], Nance-Horan, Orofaciodigital syndrome (type 3), Sturge-weber and Tricho-Rhino-Phalangeal, type1.

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In the case described here is a bizarre generalized microdontia involving the entire dentition along with mandibular mesiodens without any other apparent systemic conditions.

**Case Report**
The patient was a 12 year old boy, only child of consanguineous parents, reported to the department of pedodontics and preventive dentistry with the complaint of small teeth. Parents noted small teeth ever since the eruption of permanent teeth. No abnormalities were reported, however, in their extended family.

**Physical examination**
Physical growth was within normal limits. The patient was of normal in stature, appearance, height, and weight for his age. Upon examination of the limbs, hands, skin, hair, nails and eyes were all appeared normal. No abnormality was noted in neck, back, muscles, cranium and joints as well. Intellectual and scholastic performance was also normal. His medical history was unremarkable; no other abnormalities were noted in the history apart from the difficult delivery. The child was examined and found to be free of any gross abnormalities.

His blood profile was normal. Serum calcium, phosphorous and alkaline phosphatase levels were also normal. Endocrinological investigation was carried out to rule in or out the possibility of hormonal disorder, and the results were within normal limits.

**Intraoral examination**
The intraoral soft tissues were healthy, but the teeth were abnormal in size and shape (Figure 1 and 2). Diagnostic casts were obtained to aid in diagnosis (Figure 3). Patient was in permanent dentition, teeth present were small in size. The patient had normal occlusion with excessive spacing between the teeth. Fully erupted mandibular mesiodens was present between the central incisors. The anterior teeth lacked normal size in all dimensions. Most of the anterior teeth were “peg-shaped” without the typical variation in mesiodistal and labiolingual dimensions. Almost all the maxillary anterior teeth did not have lingual pits whereas mandibular central and lateral incisors had prominent pits on the lingual surfaces. The posterior teeth were also small and exhibited a short occlusogingival dimension. Overall, the dentition was smaller than that of the average adult (Table 1 and 2). Orthopantomogram or the Intra oral periapical radiograph could not be taken because the patient was not able to afford.

The simultaneous presence of microdontia and supernumery teeth is been reported in the Cleidocranial dysplasia, Cranio-Metadiaphyseal dysplasia, Dermodontodyplasia, Hypodontia and nail dysgenesis, Orofaciodigital syndrome type 3 and Tricho-rhinophalangeal syndrome type 1. However in this case, except for the dental abnormality in the form of generalized microdontia and the presence of fully erupted mandibular mesiodens between the central incisors were found and no other clinical features observed, therefore all the syndrome associated with the simultaneous presence of microdontia and supernumery teeth were ruled out along with Taurodontism, microdontia, and dens invaginatus as well as Distal symphalangism,
hypoplastic carpal bones, microdontia, dental pulp stones, narrowed zygomatic arch (Table 3).

A diagnosis of non-syndromic occurrence of true generalized microdontia with mandibular mesiodens was made as no systemic condition was observed. The fully erupted mandibular mesiodens was extracted under local anesthesia in order to correct midline and to facilitate the orthodontic treatment.

**Discussion**

The initiating factor or factors responsible for microdontia remain obscure. Mutation in developmental regularity genes are known to cause variety of dental defects [16].

Both genetic and environmental factors are involved in the complex etiology of microdontia. Genetic factors probably play a role in the formation of microdontia. Although the proband was the only child, the presence of consanguinity in the form of both parents being maternal first cousins could suggest recessive or polygenic inheritance.

The development of a tooth has been shown to have ectodermal, mesodermal, and neural crest contributions. The variation in size of a particular tooth arises during the period when the form of the tooth is being determined by the enamel organ and the sheath of hertwig at the bell stage of enamel organ. The determination of the form of the crown is thought to be related to different regions of the oral epithelium or to the ectomesenchyme. Studies have shown that different regions of the oral epithelium rather than the underlying ectomesenchyme are initially responsible for the shape of the crown [17]. Bones dating from the Middle Ages which were excavated at Alborg, Denmark proved evidence for generalized microdontia resulting from intrauterine growth retardation [18].

On the basis of visual documentation, the patient in the current case seems to have been more severely affected in all his teeth which exhibited aberrant morphology and all his teeth which exhibited aberrant morphology and all were smaller than normal. MEDLINE search in the English dental literature for true generalized microdontia revealed zero search results. Although child’s mother had

### Table 1 Comparison of buccolingual/labiolingual and mesiodistal crown dimensions with an anatomic average* of the right side maxillary and mandibular teeth

|                  | Right side |          |          |          |          |          |          |          |
|------------------|------------|----------|----------|----------|----------|----------|----------|----------|
|                  | Central incisor | Lateral incisor | Canine | First premolar | Second premolar | First molar | Second molar | Total |
| Maxillary        | MD         | LE       | MD       | MD       | MD       | MD       | MD       | MD       |
| Average          | 8.5        | 7.0      | 7.5      | 7.0      | 7.0      | 10.0     | 9.0      | 55.5     |
|                  | 7.2        | 6.1      | 6.4      | 6.5      | 7.7      | 10.4     | 10.9     | 55.9     |
|                  | 8.5        | 7.0      | 7.5      | 7.0      | 7.0      | 10.0     | 9.0      | 55.5     |
|                  | 7.2        | 6.1      | 6.4      | 6.5      | 7.7      | 10.4     | 10.9     | 55.9     |

### Table 2 Comparison of buccolingual/labiolingual and mesiodistal crown dimensions with an anatomic average* of the left side maxillary and mandibular teeth

|                  | Left side |          |          |          |          |          |          |          |
|------------------|-----------|----------|----------|----------|----------|----------|----------|----------|
|                  | Central incisor | Lateral incisor | Canine | First premolar | Second premolar | First molar | Second molar | Total |
| Maxillary        | MD         | MD       | MD       | MD       | MD       | MD       | MD       | MD       |
| Average          | 8.5        | 7.0      | 7.5      | 7.0      | 7.0      | 10.0     | 9.0      | 55.5     |
|                  | 7.2        | 6.1      | 6.4      | 6.5      | 7.7      | 10.4     | 10.9     | 55.9     |
|                  | 8.5        | 7.0      | 7.5      | 7.0      | 7.0      | 10.0     | 9.0      | 55.5     |
|                  | 7.2        | 6.1      | 6.4      | 6.5      | 7.7      | 10.4     | 10.9     | 55.9     |

Measurements in millimeters were taken at widest portion of clinical crown on diagnostic casts. *Anatomic average taken from Wheeler, R. C.: Textbook of Dental Anatomy and Physiology, ed. 7, Philadelphia, 1993, W. B. Saunders Company, pp. 25.
| Taurodontism, microdontia, and dens invaginatus | Cleidocranial dysplasia | Craniofemuradiophyseal dysplasia | Dermomandibular dysplasia | Hypodontia and nail dysgenesis | Orofaciodigital syndrome type 3 | Tricho-rhino-phalangeal syndrome type 1 | Distal symphalangism, hypoplastic carpal bones, microdontia, dental pulp stones, narrowed zygomatic arch |
|-----------------------------------------------|-------------------------|----------------------------------|---------------------------|-----------------------------|--------------------------------|----------------------------------|--------------------------------------------------|
| Generalized microdontia                        | Autosomal recessive     | Autosomal dominant               | Autosomal recessive       | Autosomal dominant          | Autosomal recessive            | Autosomal dominant                |                                                  |
| Taurodontism of first permanent molars         | Macropelvis             | Dry skin                         | Chromosome 4              | Thin/hyperconvex/hypoplastic nails | Autosomal recessive            | Absent/small nails                |                                                  |
| Multiple teeth with one or more dens invaginatus | Arm p                   | Frontal bossing                  | Arm p                     | Prominent occipit           | Chromosome 8                   | Microdontia                        |                                                  |
| X-linked recessive inheritance                 | Normal height           | Large fontanelle                 | Thin skin                 | Frontal bossing             | Arm q                          | Other dental abnormality           |                                                  |
| Short stature - postnatal                      | Prominent eyes          | Pigmented naevi                  | Fine hair                 | Round face                  | Normal height                  | Abnormal clinical features of the limbs |                                                  |
| Absent/small nails                             | Mandibular hypertelorism | Abnormal hair texture            | Brittle hair/trichorrhexis nodosa/pili torti | Hypertelorism                | Short stature - postnatal        | Brachydactyly                      |                                                  |
| Macrocephaly                                   | Optic nerve abnormality | Sparse/absent scalp hair - localised | Sparse/absent scalp hair - generalised | Down-sloping palpbral fissures | Decreased body hair/hypotrichosis | Irregularities of length/shape of fingers |                                                  |
| Flat occiput (brachycephaly)                  | Microdontia             | Abnormal nails                   | Absent/small nails        | Other orbital abnormality    | Decreased hair pigmentation - general | Syndactyly of fingers             |                                                  |
| Frontal bossing                                | Abnormal tooth position/malocclusion/open bite | Midface hypoplasia/flat/short midface | Thin/hyperconvex/hypoplastic nails | Paresis of ocular muscles/squint | Decreased hair pigmentation - patchy | Short foot (including brachydactyly) |                                                  |
| Wide sutures/ delayed fusion of sutures        | Missing permanent teeth/retained deciduous teeth | Micrognathia/agagnathia/retrognathia | Dysplastic/thick/thick/discoloured nails | Other eye movement disorder | Fine hair | Syndactyly (other than minimal 2nd and 3rd toes) |                                                  |
| Large fontanelle                               | Anodontia/oligodontia  | Microdontia                      | Depressed premillar region | Broad/bulbous nasal tip     | Brittle hair/trichorrhexis nodosa/pili torti | Irregular length or shape of toes |                                                  |
| Facies significantly abnormal                  | Natal/neonatal teeth    | Anodontia/oligodontia            | Midface hypoplasia/flat/short midface | Cleft soft palate/bifid uvula/submucous cleft | Sparse/absent scalp hair - generalised | Other skull abnormality             |                                                  |
| Small face                                     | Supernumerary teeth     | Supernumerary teeth              | Micrognathia/agagnathia/retrognathia | Microdontia                 | High hairline - front          | Absent/small/hypoplastic carps |                                                  |
| Hypertelorism                                  | Dental caries           | Other dental abnormality         | Absent/decreased eyebrows/lateral thinning | Abnormal tooth position/malocclusion/open bite | Thin/hyperconvex/hypoplastic nails | Symphalangism                      |                                                  |

Table 3 Comparison of conditions associated with the simultaneous presence of microdontia and supernumery teeth along with taurodontism, microdontia, and dens invaginatus as well as distal symphalangism, hypoplastic carpal bones, microdontia, dental pulp stones, narrowed zygomatic arch.
| Condition                                                                 | Table 3 Comparison of conditions associated with the simultaneous presence of microdontia and supernumery teeth along with taurodontism, microdontia, and dens invaginatus as well as distal symphalangism, hypoplastic carpal bones, microdontia, dental pulp stones, narrowed zygomatic arch. (Continued) |
Table 3 Comparison of conditions associated with the simultaneous presence of microdontia and supernumerary teeth along with taurodontism, microdontia, and dens invaginatus as well as distal symphalangism, hypoplastic carpal bones, microdontia, dental pulp stones, narrowed zygomatic arch. (Continued)

| Condition | Associated Feature |
|-----------|--------------------|
| Deafness - conductive | Absent/abnormal sinuses |
| Other hearing abnormality | Wormian bones |
| Narrow/sloping shoulder/ hypermobile shoulders | Sclerotic/hyperostotic facial bones |
| Pectus excavatum (funnel chest) | Other skull abnormality |
| Bell-shaped chest | Hyperostotic/wide clavicle |
| Thoracolumbar general kyphosis | Abnormal rib structure including fusion |
| Gibbus/localised kyphosis | Widened ribs |
| Scoliosis | Irregular shape of pubic and ischial bones |
| Hyperextensible/ hypermobile joints | Absent/hypoplastic/short femur |
| Small hand | Femora short/deformed/bowed |
| Brachydactyly | Other abnormal femur |
| Seizures of any type | Bow legs - genu varum |
| Hypotonia | Cranial nerve/nuclei |
| Imperforate anus/ anal stenosis | Mental retardation - moderate/severe |
| Horseshoe/fused/ectopic kidneys | Hypoplastic/small nostrils |
| Hypospadias/epispadias | Abnormal columnella |
| Undescended/ectopic testes | Hypotonia |
| Wilms tumour | Thin lips |
| Deafness - conductive | Movement disorder - dystonia/chorea/ tremor/spasm |
| Other hearing abnormality | EEG abnormality |
| Narrow/sloping shoulder/ hypermobile shoulders | Deeply grooved philtrum |
| Pectus excavatum (funnel chest) | Short sternum |
| Bell-shaped chest | Microdontia |
| Thoracolumbar general kyphosis | Abnormal tooth position/ malocclusion/ open bite |
| Gibbus/localised kyphosis | Supernumerary teeth |
| Scoliosis | Anteverted/prominent/bat ears |
| Hyperextensible/hypermobile joints | Long/large ear |
| Small hand | Pectus carinatum (pigeon chest) |
| Brachydactyly | Thoracolumbar general kyphosis |
| Seizures of any type | Scoliosis |
| Hypotonia | Hypertelorism/hypermobile joints |
| Imperforate anus/ anal stenosis | Small hand |
| Horseshoe/fused/ectopic kidneys | Brachydactyly |
| Hypospadias/epispadias | Clinodactyly of 5th finger |
| Undescended/ectopic testes | Terminal hypoplasia fingers |

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| Condition                                                      |
|----------------------------------------------------------------|
| Delayed skeletal maturation                                    |
| Poorly ossified calvarium/Soft skull                           |
| Absent/abnormal sinuses                                        |
| Wormian bones                                                  |
| Platybasia/basilarch impression                                 |
| Enlarged foramen magnum                                        |
| Small/absent scapula                                           |
| Winged/other abnormal scapula (See Shoulder)                   |
| Absent/ hypoplastic clavicles                                  |
| Pseudarthrosis of clavicle                                     |
| Short ribs (circumferential)                                   |
| Under-/unossified sternum                                      |
| Hypoplastic/ absent ribs                                       |
| Dorsal wedging of vertebral bodies                             |
| Narrow/trapezoid iliac wings (lack of flare)                   |
| Horizontal/flat acetabular roof                                |
| Spindle shaped/ tapered fingers                                 |
| Ulnar deviation of fingers                                     |
| Other hand abnormality                                         |
| Mental retardation of any degree                               |
| Abnormal cardiovascular structure/function                      |
| Winged/other abnormal scapula (See Shoulder)                   |
| Coxa vara                                                      |
| Cone shaped epiphyses                                          |
| Small femoral head epiphyses                                   |
| Flat femoral head epiphyses                                    |
| Deformed/ irregular femoral head epiphyses                     |
| Broad femoral neck                                              |
| Cone-shaped epiphyses of proximal phalanges                    |
| Some phalanges short and deformed                              |
| Cone-shaped epiphyses of middle phalanges                      |
| Cone-shaped epiphyses of distal phalanges                      |
Table 3 Comparison of conditions associated with the simultaneous presence of microdontia and supernumery teeth along with taurodontism, microdontia, and dens invaginatus as well as distal symphalangism, hypoplastic carpal bones, microdontia, dental pulp stones, narrowed zygomatic arch. (Continued)

| Condition Description |
|------------------------|
| Delayed ossification of pubic and ischial bones |
| Open pubic symphysis in adults |
| Coxa valga |
| Coxa vara |
| Dislocated hip |
| Cone shaped epiphyses |
| Fibulae a-/hypoplastic/under-/unossified |
| Cone-shaped epiphyses of proximal phalanges |
| Cone-shaped epiphyses of middle phalanges |
| All middle phalanges short/deformed |
| Cone-shaped epiphyses of distal phalanges |
| All distal phalanges short/deformed |
difficult delivery, it was insignificant and neither microdontia nor mesiodens has been reported in the literature. The prevalence of mesiodens varies between 0.09 and 2.05% in different studies. In permanent dentition, a 0.15 to 3.8% incidence of mesiodens has been reported [19]. Erupted supernumerary teeth in the mandible are rare, is about 0.01% which indicated marked low value [20]. Supernumerary teeth in the mandible anterior region in this case is fully erupted which is unusual.

Sexual dimorphism is reported by most authors with males being more commonly affected. Hogstrum and Andersson [21] reported a 2:1 ratio of sex distribution. A study of supernumerary teeth in Asian school children found a greater male to female distribution of 6.5:1 for Hong Kong children [22] which indicates that supernumerary teeth is more common in males than females which is consistent in our case.

Non-syndromic multiple supernumerary teeth occur most frequently in the mandible region especially premolar region followed by molar and anterior region [9]. Few cases of non-syndromic multiple supernumery teeth have been reported [23,24] however in the present case non-syndromic single supernumery tooth was observed in the mandibular anterior region.

Evidence regarding etiology of mesiodens indicates that genetic susceptibility together with environmental factors might increase the activity of dental lamina leading to formation of the extra tooth/teeth [19]. A number of theories have been proposed as regards the causes of the occurrence of supernumerary teeth: 1) Atavism theory [8,24,25] 2) Independent hyperactivity of the dental lamina [24,25] and 3) Dichotomy of the tooth bud are also suggested as a possible etiological factors [8,25]. However, none of these theories alone offers a sufficient explanation for this phenomenon.

Since mesiodens may interfere with normal occlusal development, in the present case an early diagnosis could have prevented the lower diastema formation. Early diagnosis and treatment of patients with supernumerary teeth are important to prevent or minimize complications. The patient did not show any abnormal systemic manifestations, all the syndrome associated with the dental anomalies were ruled out. The simultaneous presence of supernumery teeth and the generalized microdontia is very rare. To our knowledge, this is the first such case of non-syndromic occurrence of true generalized microdontia in association with mandibular mesiodens. Such unusual nature of dental anomaly has not been reported so far in the literature.

**Conclusion**

The dental finding seen in this case is certainly rare. The case is also sporadic, with no positive family history. The wide variation in clinical manifestations in cases of non-syndromic occurrence of dental anomalies is challenging and as an area for further research. Mesiodens are familiar to pediatric dentists and orthodontists as one of the more common anomalies to affect the developing dentition and it demands a multidisciplinary assessment.

**Consent**

Written informed consent was obtained from the patient for publication of this Case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

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**Authors’ contributions**

SB and SK drafted the manuscript paper, analysed the patient’s history and contributed to the writing of the final version as well as extracted the mesiodens. Each author reviewed the paper for content and contributed to the writing of the manuscript. All authors approved the final report.

**Competing interests**

The authors declare that they have no competing interests.

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