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Case study

Hypodontia, Microdontia and Tooth rotation: A rare clinical triad

Pushappreet Kaur*

Dental officer, Guru Nanak Dev Dental Hospital, SultanpurLodhi, Kapurthala, Punjab, India – 144626
**Corresponding author E-mail:kaurpushp@gmail.com*

Abstract

Hypodontia, microdontia and rotations are developmental dental anomalies resulting from genetic and epigenetic disturbances during tooth initiation, morphogenesis and eruption stages; with variable effects on function and dentofacial esthetics depending upon their severity. Hypodontia and microdontia may occur in syndromic or isolated non-syndromic forms based on presence or absence of congenital defects. This article briefly reviews hypodontia, microdontia and tooth rotation; and discusses a rare case presenting combination of these anomalies along with a possible treatment proposal.

Keywords:*Anomalies; Case;Hypodontia;Microdontia; Rotations*

1. Introduction

Hypodontia refers to congenital agenesis of less than six teeth excluding third molars which may affect primary and /or permanent dentition and has been generally classified into: mild (1-2 teeth missing), moderate (3-5 teeth missing) and severe (6 or more teeth missing) [Tan et al. 2011]. Microdontia (microdentism/microdontism) is a condition where affected teeth are smaller than normal i.e. outside the usual limits of variation. Neville and Shaefer [Shafer et al. 1993, Neville et al. 2009] classified microdontia chiefly into three types: True generalized, Relative generalized, Single tooth microdontia which is a common condition, frequently affecting maxillary lateral incisors (peg laterals/conical teeth) and third molars and was further classified by Bargale et al., (2011) [Bargale & Kiran 2011] as: Microdontia of the whole tooth, crown or root alone.
Hypodontia and microdontia show syndromic and non syndromic forms depending upon their association with birth defects. Non-syndromic/familial form of hypodontia occur as an isolated trait, affecting variable numbers of teeth and may appear either sporadically or in a familial fashion within a family pedigree [Tan et al. 2011], following autosomal dominant, autosomal recessive or sex-linked patterns of inheritance, with considerable variation in both penetrance and expressivity [Shimizu & Maeda 2009]. Syndromic forms of hypodontia are associated with syndromes like ectodermal dysplasia, Down's syndrome, Ellis van Crevald syndrome and birth defects like cleft lip and palate [Shimizu & Maeda 2009] where it presents as a constitutive part in combination with whole clinical spectrum [Larmour et al. 2005]. The syndromes associated with microdontia are Gorlin-Chaudhry-Moss syndrome, Williams's syndrome, Chromosome d/u, 45X [Ullrich-Turner syndrome], Hallermann-Streiff, Chromosome 13 [Trisomy 13], Orofaciodigital syndrome (Type 3), Rothmund-Thomson syndrome, Oculomandibulo-facial syndrome, Tricho-Rhino Phalangeal, Type1 Branchiooculo-facial syndrome [Bargale & Kiran 2011].

Tooth rotation is a positional anomaly characterized subjectively by any evident (at least 20°) mesiolingual or distobuccal intra-alveolar displacement of tooth around its longitudinal axis as a result of developmental changes during permanent teeth eruption. Gupta et al. in his study classified tooth rotation into three groups: <45°, 45-90° and >90° with majority of tooth rotations showing angulations between 45° and 90°, followed by <45° rotations [Gupta et al. 2011].
This article discusses above three developmental anomalies and reports few findings, part of which show rare individual presentation; while their combined occurrence has never been reported before, making this case report educationally and clinically important.

2. Case report

A healthy 21 year old male patient reported with a chief complaint of pain in the right lower quadrant which was dull and continuous. He also gave a history of heightened thermal sensitivity and pain on biting in that region. Upon clinical examination, a deep class 1 cavity with buccal extension was found with respect to 46 and the tooth was moderately tender on percussion. Besides, spaces were noticed in the anterior region which were generalized in maxillary and localized in mandibular arch, giving the patient an unaesthetic appearance (Fig. 1). Further exploration led to observation of few dental anomalies which included peg shaped (conical) 41, missing 31, 32, 12, 22 and rotated 14, 24 and 33 (Fig. 1 & 2). The space in anterior mandible was not wide enough to accommodate two missing incisors which could possibly be explained by mesial migration of distal teeth. Secondly, loss of dental midline was noticed although the patient had normal molar relationship. However, soft tissues showed normal color, consistency and contour and past dental history confirmed absence of previous trauma or extraction. Past medical and family history was also non-conclusive and no relative, distant and/or close, was reported to have similar findings. Extra-oral examination revealed normal facial profile, facial height and jaw relationship. Physical examination showed normal gait, height, weight and appearance with skin, hair, nails and eyes also appearing normal. Skeletal examination revealed absence of muscular, joint and spinal problems. Patient's intellect, hearing and speech were also normal. Blood profile was carried out and showed normal levels of serum calcium,

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phosphate and alkaline phosphate. Apart from that, exploratory IOPAR's were performed in relation to mandibular and maxillary anterior regions using bisecting angle technique; to further investigate the anomalies and rule out the possibility of impaction of missing teeth. OPG was the first preference but unavailability of this technique in our hospital and the nearest centre being couple of hours away, led to patient's refusal.



Fig. 1: Labial View of Both Arches Showing Obvious Compromisation of Esthetics.



Fig. 2: (Left) OcclusalView of Maxillary Arch Showing Generalised Spacing in the Anterior Region on Account of Missing 12 and 22; and Distolingual Rotation of 14 and 24



Fig. 3: (Right) Occlusal View of Mandibular Arch Showing Well Aligned Teeth Except for Missing 31 and 32, Mesiolingually Rotated 33 and Peg Shaped/Conical 41. Loss of Space in 31 and 32 Region Can Also Be Noticed.

Maxillary assessment was carried out by performing two IOPAR's, one with respect to 11 and 13 which revealed no sign of 12 and second with respect to 21 and 23 which revealed absence of 22 (Fig. 4 & 5). IOPAR of the mandibular anterior region was performed with respect to 41 and 33 which revealed absence of 31, 32 and abnormal shape of 41 having peg shaped crown and short root but normal pulpal and periodontal status (Fig. 6). Hence, impaction of involved teeth was also ruled out.





Fig. 4: (Left) &Fig. 5 (Middle): IOPAR of Maxillary Anterior Region Showing Agenesis of 22 (Left) and 12 (Middle).



Fig. 6: (Right) IOPAR of Mandibular Anterior Region Showing Conical Crown and Short Root of 41 as Well as Agenesis of 31 and 32.

Finally, after careful evaluation of the available data, it was concluded that patient showed only dental anomalies without any sign of underlying genetic disorder or condition. A final diagnosis of isolated non-syndromichypodontia (unilateral agenesis of mandibular incisors and bilateral agenesis of maxillary laterals) and microdontia (peg shaped mandibular central) with associated bilateral rotation of maxillary bicuspids and unilateral rotation of mandibular canine was established. Patient was informed about these anomalies and thoroughly explained about the functional and esthetical complications associated with it. Various treatment options were suggested to the patient for improvement of his esthetics and restoration of function but he sadly refused for any treat-

ment. Regarding 46, a diagnosis of chronic pulpitis was made and patient was advised root canal therapy for the same for which he reluctantly agreed. The institutional ethical board approval was obtained.

3. Discussion

Tooth development is an intricate process involving programmed sequencing of information exchange between ectodermal, mesodermal, and neural crest elements defining the regulation of initiation (tooth region and number), morphogenesis (tooth type, size, shape including dimensions and cusp number) and differentiation (tooth structure – enamel and dentine formation and mineralization) [Brook 2009]. Anomalies of size arise during morphogenesis period in bell stage while determination of tooth form by the enamel organ and the sheath of hertwig; whereas anomalies of tooth number occur in the initiation stage [Osborn & Ten Cate 1976]; and anomalies of position occur in eruption phase when a tooth moves from its area of development towards functional position [Ferriera 2004]. These development errors occur due to disturbances in the regulatory genetic and epigenetic factors with environmental factors like trauma, surgical procedures, chemotherapy, cranial irradiation early in development, somatic diseases such as syphilis, scarlet fever, rickets, nutritional disturbances during pregnancy or infancy playing possible influencial role [Vastardis 2000, Antunes et al. 2013].

According to Endo et al. [Endo et al. 2007], clinical implications associated particularly with agenesis of mandibular incisors include disturbance in tongue-lip pressure balance, lack of lingual support and severe malocclusion; usually class II Div I malocclusion along with severe anterior deep bite, absence of dental midline and variable large space in the anterior region. Occasional association of Class III malocclusion is also noted [Endo et al. 2006]. This patient presented absence of dental midline and large space in the lower anterior region resulting in an unaesthetic appearance. However, molar relationship appeared normal.

Majority of hypodontia cases (approx. 80%) reported only 1 or 2 missing teeth with few cases (10%) showing 4 or more missing teeth and only fewer (>1%) presenting 6 or more missing teeth [Larmour et al. 2005]. This patient had four permanent missing teeth (moderate hypodontia) which is relatively less frequent scenario.

Many authors stated strong association of microdontia with hypodontia [Shafer et al. 1993, Altug-Atac & Erdem 2007, Anziani et al. 2010]. Additionally, family studies have indicated that peg shaped upper lateral incisors; impacted canines, rotated bicuspids, and short root anomaly are caused by the same genetic elements responsible for agenesis of incisors and premolars [Baccetti 1998]. All these evidences point towards the possibility of potential genetic interlinking between these anomalies; and our patient showing combination of hypodontia, microdontia and rotations eloquently advocate this prospect.

Numerous studies demonstrate higher frequency of hypodontia and microdontia in females [McKeown et al. 2002, Neville et al. 2004, Larmour et al. 2005]. Contrarily, few authors noted that anomalies were more frequent in male patients than in females [Sisman et al. 2007, Tallon-Walton et al. 2010]. This case was evident of the less frequent gender in prevalence of dental anomalies. On the other hand, literature holds scant evidence regarding gender predisposition on tooth rotations.

Various reports support that frequency of hypodontia follows: Second mandibular premolar (40% to 50%), maxillary lateral incisor (25%), maxillary second premolar (20%) and mandibular central incisor (6.5%) [Rose 1966]. Regarding microdontia, four different studies conducted on Indian population showed a prevalence rate of 0.16%, 1%, 2.58% and 4.3% with maxillary laterals incisors (peg laterals) most frequently affected [Sharma & Singh 2014]. A comprehensive search of English articles showed only six reported cases of peg shaped mandibular central incisor which included Sharma, Chanchala and Nandlal, Anziani et al., Ramachandra et al., Malleshi et al., and Shalini and Sudeep [Sharma 2001, Ramachnadra et al. 2009, Anziani et al. 2010, Chanchala&Nandlal 2012, Malleshi et al. 2014, Sharma & Singh 2014]. Above mentioned frequency reports of various teeth affected by hypodontia and microdontia clearly indicate the rarity of this case.

A study reported that mandibular second premolars were most frequently rotated teeth followed by mandibular first premolars and maxillary central incisors, which showed similar prevalence rate [Gupta et al. 2011]. However, another study conducted by Teixeira, Martins, Lascala et al. [Teixeira et al. 2008] observed lower canines as most commonly rotated teeth with lower incisors showing low frequency.

The overall prevalence rate of hypodontia, microdontia and tooth rotations range from 1.6% to 13.3% [Anziani et al. 2010], 0.8% to 8.4% [Neville et al. 2005] and 2.1-5.1% [Shpack et al. 2007] respectively among different ethnic groups, with Indian population showing 4.19% hypodontia rate [Gupta et al. 2007]. Medina stated that while bilateral agenesis usually affects maxilla, the mandible mostly shows unilateral agenesis. On the contrary, few other studies reported that mandibular second premolar could be the most common symmetrically missing tooth, followed by maxillary second premolar or maxillary lateral incisor [Rakshan 2015]. However, the findings in this case were consistent with Medina study.

3.1. Treatment options

Various factors which influence treatment strategy are: severity of the defect, age of patient, financial status, mental and physical health conditions, and patient compliance. An ideal treatment plan should consider these factors and use a combined or multidisciplinary approach to target all the anomalies simultaneously. The case in this report shows three types of anomalies and requires a systematic method for its management. A possible treatment option has been proposed below.

3.1.1. Space optimization

Mandibular arch shows localized space in the anterior region whereas maxillary arch displays generalized spacing. It is important to obtain sufficient space in maxillary arch for bilateral rehabilitation of lateral incisors whereas preexisting space in mandibular arch is optimal for rehabilitation of at least one incisor. This space gain can be achieved only with the help of fixed appliance to carry out necessary tooth movements:

- Alignment of teeth in both arches
- De-rotation of 14, 24 and 33
- Retraction of 13 and 23
- Closure of midline diastema

3.1.2. Space utilization

The obtained space can now be utilized to rehabilitate 12, 11 and 31/32 with following three main methods:

- Implant
- Fixed partial prosthesis
- Removable partial denture

If mandibular space is restored using an implant, then a metal ceramic crown would be required to improve the esthetics of peg shaped 41. And if, a removable partial denture is preferred, a crown should be planned for 41 or it should be extracted followed by restoration of resultant space with acrylic removable denture. However, the patient and his family were not much interested in the treatment of anomalies and projected with an idea of cheap, quick and easy fix for all the defects. Only treatment option which was possible considering their reluctancy, included rehabilitation of localized space in lower anterior region with removable or fixed prosthesis, either with or without extraction of 41. Although, this would have meant ignoring the generalised spaces in anterior maxillary region and leaving rotated maxillary bicuspids as such but then again, it would have at least provided mild improvement of esthetics and function. However, in the end, patient simply

refused for any treatment except for 46. Possible reasons for patient's refusal may include:

1) Lack of awareness
2) Financial disability
3) Difficulty in attending the orthodontic appointments; travelling time might be a factor as the patient belonged to a distant rural area
4) Negligence
5) Hesitation regarding length of orthodontic treatment

4. Conclusion

It was important to document this case because it presented as a clinical triad of following dental anomalies:

1) Moderate hypodontia: bilateral agenesis of maxillary laterals and unilateral agenesis of mandibular central as well as lateral incisor.
2) Localised microdontia: Mandibular peg shaped central incisor.
3) Bilateral distolingual rotation of maxillary first premolars and unilateral mesiolingual rotation of mandibular canine.

A wide search of English literature yielded no such case which showed simultaneous presence of these anomalies, thus making it a rarity. Also, peg shaped mandibular central, unilateral agenesis of mandibular incisors and rotated maxillary bicuspids are rare individual findings. Such cases subtly address the proposition of close genetic linkage between different dental anomalies; thus serving as pivots that eventually assist in advancing our knowledge and guiding us in constructing an effective treatment plan. They also form valuable aids in assessing evolutionary structural and morphological changes in human dentition, and further studies should be encouraged.

5. Patient consent

Patient was informed about the intent of this article and he gave his written consent.

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Conflict of Interest

None.

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