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An Unusual Dental Anomaly in a Hypodontia Patient

Abstract: Hypodontia is the most common developmental dental anomaly known in humans. It is often associated with other dental anomalies such as disturbances in eruption, peg lateral incisors and taurodontism. This case report describes a ten-year-old female with hypodontia and a very unusual conical-shaped mandibular right central incisor, which has not previously been reported in the literature. The case highlights the importance of prompt referral to an interdisciplinary team so joint decisions on management can ensure an optimal outcome with the corresponding improvement in quality of life.

Clinical Relevance: Hypodontia and other developmental dental disorders are common and their early recognition and prompt referral is important.

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Hypodontia is the agenesis of one or more teeth in the developing dentition. With a prevalence in the permanent dentition of 1.6% to 9.6% (excluding third molars), it is the most common developmental dental anomaly known in humans.[1,2] The prevalence varies between studies owing to differences in sampling and diagnostic techniques, as well as racial variations.[2] It can be associated with certain syndromes such as ectodermal dysplasia, Down's syndrome and conditions such as cleft lip and palate.[3] Hypodontia is more common in females than males[3] by a ratio of 3:2.[4] The most commonly affected teeth in Caucasians (excluding third molars) are maxillary lateral incisors and second premolars.[1]

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Dental anomalies associated with hypodontia include the following:

- Disturbances in exfoliation and eruption;
- Spacing;
- Microdontia;
- Taurodontism; and
- An overall delay in dental development.[5]

Most of the additional anomalies are associated with the absence of second premolars (63.6%) compared to absent lateral incisors (38.4%).[1]

Microdontia can loosely be defined as a tooth that is smaller than expected. A more specific definition offered by Kaplan[6] is a small tooth with greater than 3.5 standard deviation below the sex-specific mean mesio-distal tooth size. Microdontia can encompass different morphologies, including peg-shaped teeth; these are anterior teeth in the primary or permanent dentition with a crown diameter that decreases markedly from cervical margin to incisal edge,[7] resulting in the characteristic peg shape.

As with hypodontia, the prevalence of microdontia varies between studies and racial groups.[8] In a British study, microdontia of the permanent dentition was found in 1.9% of males and 3.1% of females.[9] A study of a Japanese population found a prevalence of 1.9%, and a prevalence of 3.2% for peg-shaped teeth in the permanent dentition,[7] the maxillary lateral incisor being the most common peg-shaped tooth. The same study reported that there were no cases of peg-shaped mandibular central incisors and 0.1% cases where this tooth was microdont.[7] A Turkish study of 3043 patients found a prevalence of microdontia of 1.58% and there were no peg-shaped mandibular central incisors and only two cases of peg-shaped mandibular lateral incisors.[8]

As tooth anomalies affecting mandibular permanent central incisors are rare, this paper describes an unusual case of a conical mandibular right central incisor and microdont mandibular left central incisor in a patient with hypodontia.

Case report

A ten-year-old female patient was referred to the interdisciplinary hypodontia clinic of a UK Dental Hospital. Her general health was good and her hair and nails normal in appearance. There was no history of hypodontia of the primary dentition and no family history of missing teeth. The patient is an only child.

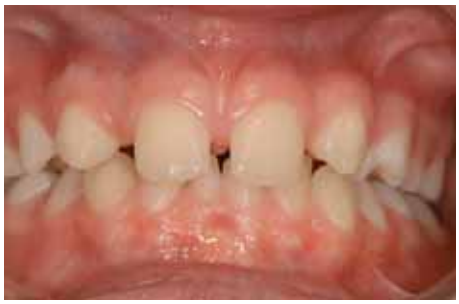

Figure 1. Anterior view in occlusion.

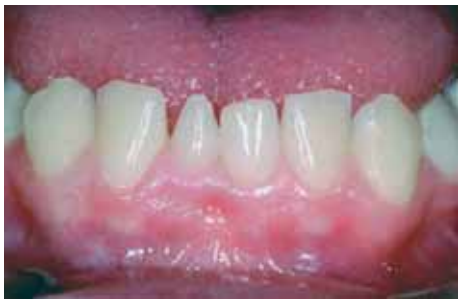

Figure 2. Mandibular anterior view.

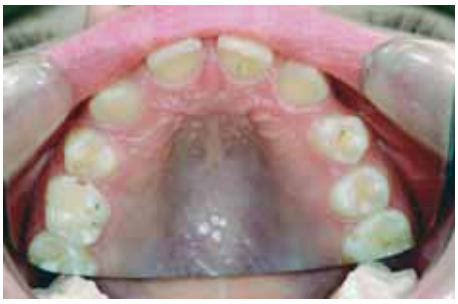

Figure 3 Maxillary arch.

Figure 4. Mandibular arch.

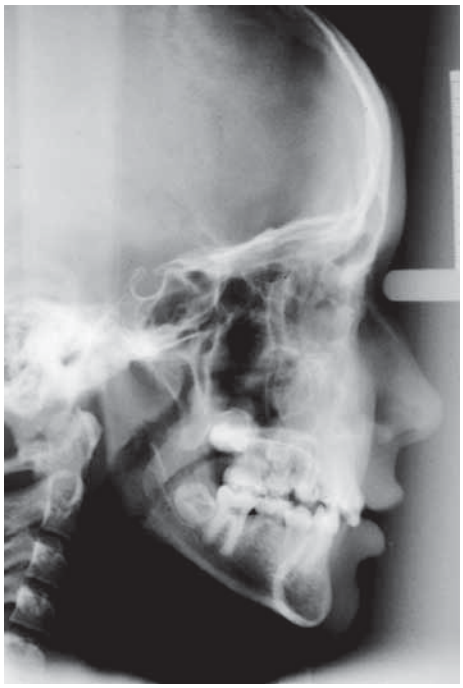

Figure 5. Lateral ceph.

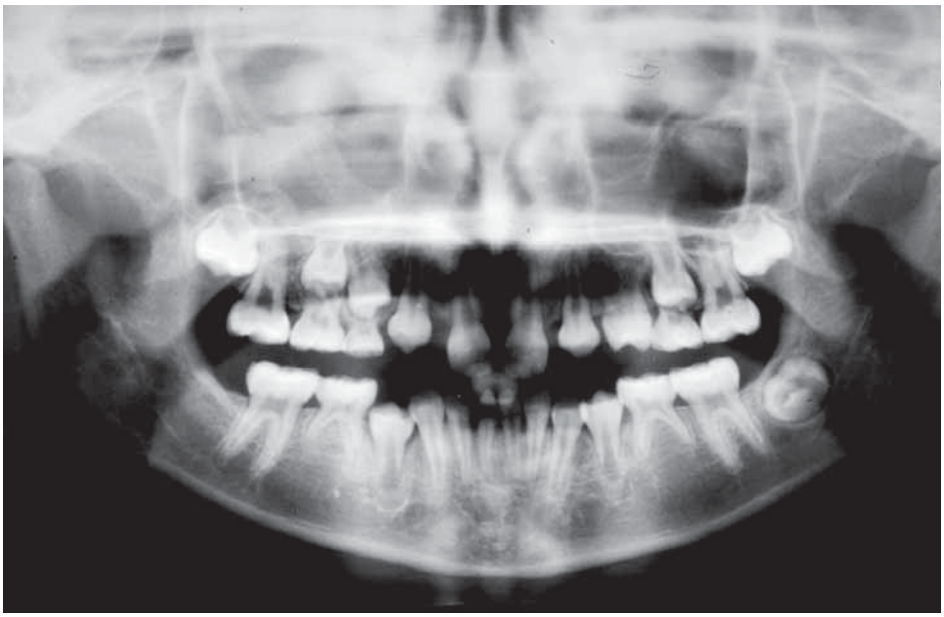

Figure 6. Orthopantomogram.

Clinical examination revealed the patient to have facial symmetry and to be in the mixed dentition. The following teeth were erupted (Figures 1–4):

- UR6, URE, UR4, UR3, UR1/UL1, UL3, UL4, UL5, UL6
- LR6, LRE, LR4, LR3, LR2, LR1/LL1, LL2, LL3, LL4, LLE, LL6

Caries was present in the maxillary right second primary molar (URE). The permanent teeth were caries free and oral hygiene was good. The mandibular central incisors were microdonts and the mandibular right central incisor conically shaped (Figure 2).

Orthodontic examination revealed a Class I antero-posterior relationship with normal vertical proportions (Figure 5). The incisal relationship was Class I with an overjet of 4 mm and overbite of 80%. There was mild mandibular arch crowding and the maxillary arch was spaced. No crossbites were present.

Clinical and radiographic examination revealed the following teeth to be congenitally absent as listed below (Figure 6):

- UR8, UR2/UL2, UL8
- LR8, LR7, LR5/LL5, LL8

It is important to take an interdisciplinary approach to the management of children with missing teeth in order to ensure the correct timing of intervention for each specialty at all stages of treatment. The principal aim in the management of hypodontia cases is to replace missing teeth to improve the patients' appearance, speech and mastication.[1] It is also important to manage existing disease and prevent further caries and periodontal disease.

Other than the restoration of the maxillary right second primary molar, this patient required no immediate dental care as the rest of her dentition was reasonably well maintained and she attended her GDP regularly. She had no particular concerns regarding her appearance at the time of presentation. Preventive advice was given and the patient was to be followed up by the GDP.

The treatment plan for the

patient (at an appropriate age) was for the use of a palatal implant for anchorage in conjunction with fixed appliances to distalize the maxillary buccal segments and open the lateral incisor spaces. These spaces will initially be restored with acid etch bridges with a view to possible implants in the future. In the mandibular arch the second primary molars will be gradually reduced in size and fixed appliances used to align the teeth prior to building up the conical mandibular right central incisor using composite resin. Her treatment was to commence when she was around 12 years of age in order to allow further maturation of the dentition.

Discussion

This case highlights the association between hypodontia and other dental anomalies that potentially complicate management of these patients. In all cases of developmental dental anomalies, early recognition is important and correct referral to a specialist team is vital to attain the best outcome for the child.

The outcomes of treatment can be measured with regard to changes in quality of life. It is recognized that conditions such as hypodontia and clefting can adversely affect quality of life.[10,11] The number of teeth present affects eating habits from both functional and social perspectives.[1]

It is important to recognize and convey to the patient and family that treatment of complex cases requires a long-term commitment from the patient and family to attend multiple appointments and undergo treatment that takes several years. This can be achieved by good communication between all interdisciplinary team members and the parents/carers of the child. The use of Kesling set-ups is not only vital for treatment planning but can aid communication of the final aims of the treatment to the child and family.

The use of palatal implants as an alternative to conventional anchorage is increasing. It has been shown that patient acceptance is excellent and they avoid problems associated with the use of headgear, such as risk of facial injury and compliance.[12] Endosseous implants do not move when a force is applied to them and therefore can be used to reinforce anchorage.[12] They have been shown to be as good as conventional headgear in a randomized control trial of 47 patients.[13] The planning stages are extremely important to ensure optimal three-dimensional positioning. There have been problems reported with a high rate of failure of palatal implants. One study[12] had 3 out of 20 palatal implants which failed to osseointegrate, another[13] had 6 out of 24 which failed to gain primary stability at the first attempt. Reasons for this may be related to variations in bone quality of the palate, the mid-palatal suture and the surgeon's experience.[12,13]

There are many explanations and theories as to the causes of tooth agenesis and variations in size and morphology. Familial trends are common[9] and provide a basis for the work of molecular geneticists investigating the genetic background of these variations. Monogenic modes of inheritance have been confirmed by genetic mapping of affected families. Vastardis[14] identified a point mutation on chromosome 4p responsible for tooth agenesis in one family. The culprit gene was discovered to be MSX1. This gene has previously been identified as important in craniofacial development and conditions such as cleft lip and palate.[15] Tooth development is a very complex process and involves many genes in addition to MSX1.[14] Other suggestions for mode of inheritance include autosomal recessive, sex-linked and polygenic patterns.[5]

Grahen[16] concluded that, in most cases, hypodontia is mainly determined by autosomal dominant trait with incomplete penetrance and variable expressivity, whereas peg teeth and unilateral hypodontia represent incomplete expression due to reduced penetrance.

Another proposition is that there is a multifactorial model for tooth size based on a continuous distribution, with thresholds determining hypodontia and supernumeraries. Brook found that crown dimensions in patients with hypodontia were significantly smaller than controls and that the whole dentition was affected.[9] The severity of the hypodontia was related to the degree of difference in crown size. The degree of crown taper from gingival margin to incisal edge increased in relation to severity of hypodontia in certain teeth. Tooth dimensions were also found to be smaller in the non-hypodontia first degree relatives of those with severe hypodontia.

An anatomical cause for tooth agenesis is suggested by Svinhufvud *et al.*[17] Certain regions during tooth development are more susceptible to factors influencing agenesis, so called 'fragile sites' more susceptible to epigenetic influences. These sites include the maxillary lateral incisor, where the median nasal and lateral maxillary processes fuse, and mandibular incisors at the mandibular symphysis. Another 'fragile site' is the distal end of the primary dental lamina which is the site of the second premolar but, interestingly, only the permanent dentition is affected.

Evolutionary trends towards smaller jaws has also been cited.[5,8,18] The number of teeth diminishes in parallel with the changes in jaw skeleton.[18] Clayton[19] observed it was usually the last tooth in the series that was missing, leading him to hypothesize that these missing teeth were 'vestigial organs'. These were of no practical value and offered no selective advantage, resulting in their loss.

Environmental factors, such as infections during foetal life and early childhood, are also cited as causes of hypodontia and microdontia.[18] This can include syphilis, rickets and irradiation of the jaws during odontogenesis.

Conclusion

This case highlights the association between hypodontia and other dental defects which can affect management and treatment planning. The importance of early referral to an interdisciplinary team is emphasized. In the case of this child, she was referred at an early age and, with co-operation from her and her family, a favourable outcome will be achievable with the associated improvements in quality of life.

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