CLINICAL SECTION

Ectopia or concomitant hypohyperdontia? A case report

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This report describes the unusual appearance seen on a panoramic radiograph of an orthodontic patient which the authors argue may represent ectopia or concomitant hypohyperdontia of the mandibular premolar teeth.

A literature review describes the frequency of such anomalies in this area from previous studies.

The presenting features of the patient and the differential *diagnoses* are explored. Treatment planning is discussed and treatment carried out in this particular case is detailed.

The unusual symmetrical bilateral anomalies in this patient may point to a genetic determinant of tooth germ position and/or movement.

Key words: Ectopia, hypohyperdontia, tooth germ, premolar, eruption

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Introduction

Little, if anything is known about the mechanisms which determine pre-eruptive tooth mechanisms. The tooth germ of the mandibular second premolar usually develops within a crypt finally positioned between the roots of the deciduous second molar.[1] The unusual appearance in the mandibular premolar area noted on a radiograph of an orthodontic patient (Figures 1 and 2) prompted debate on whether the clinical situation represented an ectopic position of development of the second premolar crypts or hypodontia of the second premolars with concomitant supplemental first premolars.

Second Premolar Ectopia

Ectopic second premolar teeth are a well-recognized anomaly. It was noted by Stafne[2] that migration of unerupted teeth was most often seen in premolars, then canines and third molars. The mandibular second premolar is more commonly involved than the first. Although the exact mechanism of migration of the tooth is ill-understood it appears that the direction of movement is related to the movement of the crown and is that of least resistance through the medullary spaces of the jaw bone. Normal eruption occurs after the formation of the crown of the tooth and is guided by the gubernacular cord towards the oral cavity.[3] Obviously any local or systemic abnormality, which interrupts the continuity of the cord, may alter the path of tooth eruption.

Sutton,[4] who reviewed sixty-two cases of unerupted mandibular premolars, found that 93.5% migrated in a distal direction. It was hypothesized that this was related to the initial distal angulation of the unerupted tooth and the frequent early loss of the first molar.

The authors are unaware of any population studies relating to the prevalence of ectopic mandibular second premolars.

Second Premolar Hypodontia

The phenomenon of hypodontia of second premolar teeth is well recognized. Data from Caucasian populations in North America, Australia and Europe were included in a recent meta-analysis.[5] The 95% confidence interval for agenesis of this tooth in 48,274 persons was between 2.91 and 3.22% making this the most frequently absent in the adult dentition. This is in agreement with another recent meta-analysis.[6] Hypodontia has been demonstrated as an autosomally inherited dominant condition with varying expression and incomplete penetrance.[7,8] In addition, it has been found that congenitally absent second premolars are significantly related to cleft lip, cleft palate and cleft lip and palate.[9]

Supernumerary Premolars

A number of papers examining the prevalence of dental anomalies note a lower prevalence of supernumerary

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Figure 1 (a-g) Facial and intra-oral views of patient (20-04-99)

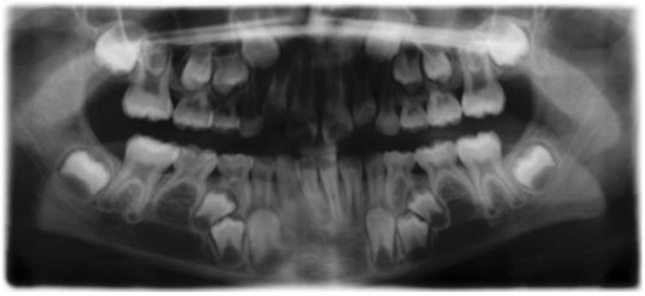

Figure 2 Panoramic radiograph (20-04-99)

teeth than other developmental dental abnormalities. In a survey of 2,000 schoolchildren,[10] Brook found that supernumerary teeth were present in 2.1% of permanent dentitions. Males are affected approximately twice as frequently as females in the permanent dentition.[11] Of these, most commonly occurring is the mesiodens. Supernumerary premolars have been reported[12] to represent around 8.0% of all supernumerary teeth. Thus, prevalence of supernumerary premolars in a general population is low.

The aetiology of supernumerary teeth is not completely understood.

A hypothesis, well supported in the literature, is the hyperactivity theory, which suggests that supernumeraries are formed as a result of local, independent, conditioned hyperactivity of the dental lamina.[13] Supernumeraries are a recognized finding in association with palatal clefts.[14] Baccetti[15] in a controlled study found that unlike other tooth abnormalities which may be significantly linked with other dental anomalies, supernumerary teeth appear to be a separate pathologic entity.

Concomitant Hypohyperdontia

Concomitant hypohyperdontia is the conveniently concise term introduced by Camilleri[16] to describe the simultaneous presence of hypodontia and supernumerary teeth. Other case reports describe this phenomenon.[17–19] This is a very rare dental anomaly and there are no published estimates of prevalence of which the authors are aware. Aetiology is obscure and as asserted by Baccetti,[15] the two occurrences are probably unrelated phenomena. Interestingly though, two papers report cases in patients with Down syndrome.[17,18] To the authors' knowledge, no case has been reported describing bilateral hypohyperdontia in the mandibular premolar region.

Case History

The patient, a female aged eight years and two months (Figure 1a,b), was referred by her general dental practitioner concerned by the slow eruption of the upper right lateral incisor.

Examination took place in Dundee Dental Hospital.

The child's medical history was unremarkable reporting eczema and a possible allergy to penicillin. Both child and parent were unaware of any trauma to the teeth and jaws or any perinatal infection suffered by child or mother. In addition, there was no family history of dental anomalies in the child's parents or siblings.

Extra-orally the patient appeared normal. Clinical examination showed the child to be in the mixed dentition stage with the following teeth present (Figure 1b–g):

Upper arch: UR1, URC, URD, URE and UR6
UL1, UL2, ULC, ULD, ULE and UL6
Lower arch: LR1, LR2, LRC, LRD, LRE and LR6
LL1, LL2, LLC, LLD, LLE and LL6

Oral hygiene was fair and caries was noted in the lower right deciduous molars.

The patient had a Class I skeletal pattern with a Class I incisor relationship. The lips were competent. The overjet was measured as 4 mm and the overbite at 3 mm. Molar relationships were class one on the left and a half unit class two on the right. The lower right lateral incisor was lingually displaced and rotated and deciduous second molars were in cross-bite on the right side, but there was no detectable mandibular displacement on closure.

Space analysis from the mesial of the first permanent molar to the distal of the lateral incisor showed 21.5 mm on the lower right hand side and 22 mm on the lower left hand side. In the upper arch, 22 mm was measured on both the left and right hand side.

A panoramic radiograph (Figure 2) revealed UR2 to be present and in an eruptive position. A maxillary anterior occlusal view was also exposed to aid with localisation of the canine teeth with the parallax technique. This confirmed their buccal eruptive path. The panoramic radiograph also confirmed the presence of all other teeth with the exception of third molars. Of great interest was the incidental finding that the lower second premolars seemed to be ectopically positioned. Radiographically the lower second premolars appeared to be erupting mesially and resorbing the roots of the lower deciduous first molars. In addition the lower first premolars were vertically impacted beneath the aberrantly erupting second premolars.

The diagnosis was one of bilateral ectopic mandibular second premolars causing pathological resorption of the roots of the lower deciduous first molars and impaction of the lower first premolars. These factors were complicating normal development in the early mixed dentition.

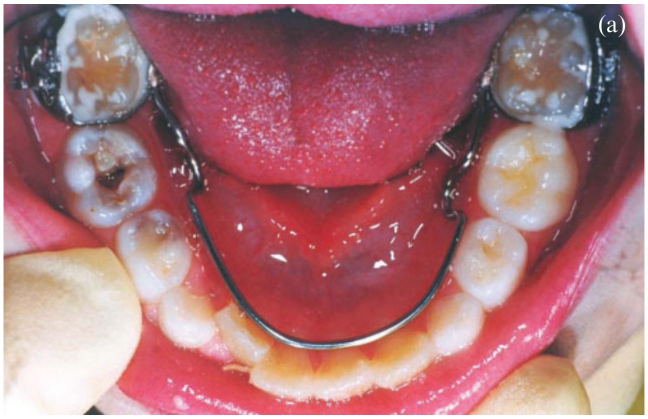

Figure 3 (a) Intra-oral views of space maintainer prior to extractions (14-05-99) and (b) approximately 15 months later (05-09-00)

Treatment objectives

Relief of crowding.

Sufficient space existed in the arch to accommodate all the premolar teeth.

Crowding was significantly increased in the lower first premolar region. It was proposed that all the lower deciduous molars be removed to allow the premolars to erupt unimpeded and then to consider the treatment options at that point.

Maintenance of space

With extraction proposed to relieve crowding in the lower premolar areas it was important to maintain space for eruption of premolars and subsequent orthodontic treatment. The rationale for this was to simplify or obviate 2nd-phase orthodontic treatment.

Treatment Sequence

Impressions were taken to provide a space maintainer. This was constructed with a lingual arch welded to two orthodontic bands which were cemented to the lower first molars on 14th May 1999 with a glass ionomer cement (Figure 3a).

The lower right deciduous molars were extracted under local anaesthetic on 19th May 1999 but due to poor patient compliance the lower left deciduous molars were extracted under general anaesthetic on 14th June 1999. Both were uneventful.

The patient was generally unhappy with any kind of dental intervention, and while willing to wear a space maintainer was not prepared to have further orthodontic intervention. Eruption was monitored periodically and photographs (Figure 3b) plus a further panoramic radiograph were taken in September 2000 (Figure 4), with second premolars appearing first, (both visible in February 2002 around the time of the patient's 11th birthday), followed approximately 7 months later by spontaneous eruption of right first premolar. It was a further 6 months before the left first premolar appeared. When both lower premolars erupted fully, they were in reasonably good alignment, but LL4 was rotated by about 30°. Final records, including photographs (Figure 5a–g) and a panoramic radiograph (Figure 6) were taken in September 2003.

Discussion

One of the first steps in examining the paediatric dental patient is to determine the presence or absence of unerupted teeth. In this case, panoramic radiography showed that lower second premolars were ectopic.

With changes in guidelines for radiography for orthodontic patients,[20] it is unlikely that this patient would have received a panoramic radiographic examination today. Exposure to diagnostic x-radiation is carefully controlled in contemporary medical practice and the authors support these changes. It is

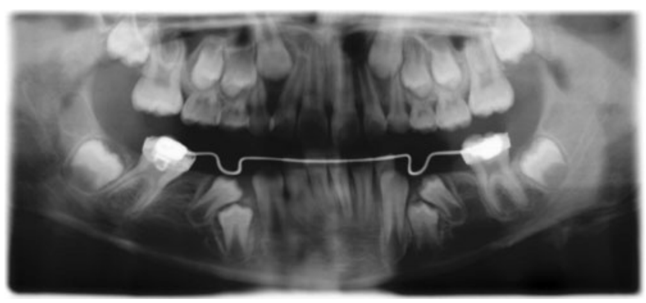

Figure 4 Panoramic radiograph (05-09-00)

Figure 5 (a-g) Facial and intra-oral views of patient (23-09-03)

interesting to note however that management in this case may have been complicated by late diagnosis of the anomaly.

Of interest to the authors was the unusual location of the ectopic second premolars on the radiograph. As most studies have shown a distal migration[4] of these

Figure 6 Panoramic radiograph (23-09-03)

teeth, bilateral mesial migration would not be expected. In the absence of any obvious local or systemic factors, it is hypothesized that this may either represent an ectopic position of the development of the second premolar tooth germ or aberrant pre-eruptive movement. Very little, if anything is known of the pre-eruptive movements of the teeth and the factors which control these. In the authors' opinion, if these represent ectopic premolars, then their unusual bilateral symmetry might suggest a genetic determinant of pre-eruptive movement or position of the tooth germ.

The fact that the lower second premolar is the tooth most frequently affected by agenesis in the buccal segment[5] and is not uncommonly ectopic,[21] is likely to have a developmental and genetic explanation. The dentition is patterned by overlapping tooth-specific domains of odontogenic homeobox gene expression in facial mesenchyme.[22] A member of the distal-less gene family (Dlx2), is expressed in the first branchial arch, and has been shown to be involved in the patterning of the murine dentition.[23] Certain signalling molecules such as Fibroblast Growth Factors (eg FGF8) control distal-less gene expression and the epithelial-mesenchymal interactions necessary for normal odontogenesis.[24] Therefore it is plausible that mutations in such genes in the human dentition will affect odontogenesis, and relative instability in the premolar domain might be explained by the cross-over of molar and canine fields.

The very unusual presentation of this case also led the authors to consider that the first premolars eruption was impeded by supplemental premolars with concomitant hypodontia of the second premolars. Whilst literature would suggest this anomaly is very rare, the lack of evidence radiographically to show any relation between the roots of the second deciduous molar and the ectopic premolar tooth germ might not make this explanation so unreasonable. In addition, what we presume to be the second premolar is erupting ahead of the first. This is unusual as in normal dental morphogenesis the first premolar chronologically precedes the second both in development and eruption.[25] The hypothetical aetiopathogenesis however did not change treatment planning in this instance.

References

1. Ten Cate AR. Physiologic tooth movement. In: *Oral histology: development, structure, and function*. 4th edn. St. Louis: Mosby; 1994, pp. 289–314.
2. Stafne EC, Gibilisco JA. *Oral roentgenographic diagnosis*, 4th edn. Philadelphia; London: Saunders; 1975.
3. Cahill DR. Histological changes in the bony crypt and gubernacular canal of erupting permanent premolars during deciduous premolar exfoliation in beagles. *J Dent Res* 1974; **53**(4): 786–91.
4. Sutton PR. Migrating nonerupted mandibular premolars: a case of migration into the coronoid process. *Oral Surg Oral Med Oral Pathol* 1968; **25**(1): 87–98.
5. Polder BJ, Van't Hof MA, Van der Linden FP, Kuijpers-Jagtman AM. A meta-analysis of the prevalence of dental agenesis of permanent teeth. *Community Dent Oral Epidemiol* 2004; **32**(3): 217–26.
6. Mattheeuws N, Dermaut L, Martens G. Has hypodontia increased in Caucasians during the 20th century? A meta-analysis. *Eur J Orthod* 2004; **26**(1): 99–103.
7. Arte S, Nieminen P, Pirinen S, Thesleff I, Peltonen L. Gene defect in hypodontia: exclusion of EGF, EGFR, and FGF-3 as candidate genes. *J Dent Res* 1996; **75**(6): 1346–52.
8. Arte S, Nieminen P, Apajalahti S, Haavikko K, Thesleff I, Pirinen S. Characteristics of incisor-premolar hypodontia in families. *J Dent Res* 2001; **80**(5): 1445–50.
9. Buenviaje TM, Rapp R. Dental anomalies in children: a clinical and radiographic survey. *J Dent Child* 1984; **51**(1): 42–46.
10. Brook AH. Dental anomalies of number, form and size: their prevalence in British schoolchildren. *J Int Assoc Dent Child* 1974; **5**(2): 37–53.
11. Kinirons MJ. Unerupted premaxillary supernumerary teeth. A study of their occurrence in males and females. *Br Dent J* 1982; **153**(3): 110.
12. Nazif MM, Ruffalo RC, Zullo T. Impacted supernumerary teeth: a survey of 50 cases. *J Am Dent Assoc* 1983; **106**(2): 201–04.
13. Liu JF. Characteristics of premaxillary supernumerary teeth: a survey of 112 cases. *J Dent Child* 1995; **62**(4): 262–5.
14. Vichi M, Franchi L. Abnormalities of the maxillary incisors in children with cleft lip and palate. *J Dent Child* 1995; **62**(6): 412–17.

15. Baccetti T. A controlled study of associated dental anomalies. *Angle Orthod* 1998; **68**(3): 267–74.
16. Camilleri GE. Concomitant hypodontia and hyperodontia. Case report. *Br Dent J* 1967; **123**(7): 338–39.
17. Acerbi AG, de Freitas C, de Magalhaes MH. Prevalence of numeric anomalies in the permanent dentition of patients with Down syndrome. *Spec Care Dentist* 2001; **21**(2): 75–78.
18. Chow KM, O'Donnell D. Concomitant occurrence of hypodontia and supernumerary teeth in a patient with Down syndrome. *Spec Care Dentist* 1997; **17**(2): 54–57.
19. Sharma A. A rare non-syndrome case of concomitant multiple supernumerary teeth and partial anodontia. *J Clin Pediatr Dent* 2001; **25**(2): 167–69.
20. Isaacson KG, Thom AR (Eds). Orthodontic Radiographs Guidelines (Guidelines for the Use of Radiographs in Clinical Orthodontics). British Orthodontic Society, August 2001.
21. Wasserstein A, Brezniak N, Shalish M, Heller M, Rakocz M. Angular changes and their rates in concurrence to developmental stages of the mandibular second premolar. *Angle Orthod*. 2004; **74**(3): 332–36.
22. Thomas BL, Sharpe PT. Patterning of the murine dentition by homeobox genes. *Eur J Oral Sci*. 1998; **106** Suppl 1: 48–54.
23. Tucker AS, Markham HJ, Green P, Doherty P, Sharpe PT. A novel approach for inhibiting growth factor signalling in murine tooth development. Inhibition of FGF's. *Eur J Oral Sci.* 1998; **106** Suppl 1: 122–25.
24. Thomas BL, Liu JK, Rubenstein JL, Sharpe PT. Independent regulation of Dlx2 expression in the epithelium and mesenchyme of the first branchial arch. *Development.* 2000; **127**(2): 217–24.
25. Eskeli R, Laine-Alava MT, Hausen H, Pahkala R. Standards for permanent tooth emergence in Finnish children. *Angle Orthod* 1999; **69**(6): 529–33.