Tooth agenesis in patients referred to an Irish tertiary care clinic for the developmental dental disorders


Précis
Hypodontia is the most common developmental disorder affecting teeth, and the mandibular second premolar is the most frequently absent tooth type.

Abstract
Purpose: This study was carried out to determine the prevalence, severity and pattern of hypodontia in Irish patients referred to a tertiary care clinic for developmental dental disorders.

Materials and methods: Details of 168 patients with hypodontia referred during the period 2002-2006 were entered in a database designed as a national record. Tooth charting was completed using clinical and radiographic examinations. The age of patients ranged from 7-50 years, with a median age of 20 years (Mean: 21.79; SD: 8.005).

Results: Hypodontia referrals constituted 65.5% of the total referrals. Females were more commonly affected than males with a ratio of 1.3:1. The number of referrals reflected the population density in this area; the majority were referrals from the public dental service. Mandibular second premolars were the most commonly missing teeth, followed by maxillary second premolars and maxillary lateral incisors; maxillary central incisors were the least affected. Symmetry of tooth agenesis between the right and left sides was an evident feature. Slightly more teeth were missing on the left side (n = 725) than on the right side (n = 706) and in the maxillary arch (n = 768) as compared to the mandibular arch (n = 663). Some 54% of patients had severe hypodontia with more than six teeth missing; 32% had moderate hypodontia, with four to six teeth missing. The most common pattern of tooth agenesis was four missing teeth.

Conclusion: Hypodontia was a common presentation in a population referred to this tertiary care clinic. The pattern and distribution of tooth agenesis in Irish patients appears to follow the patterns reported in the literature.

Introduction
Hypodontia is the term used to describe the developmental absence of one or more primary or secondary teeth, excluding the third molars. It is the most common developmental dental anomaly and can be challenging to manage clinically. Generally, hypodontia refers to the condition where there is absence of one or a few teeth only. Oligodontia is the term usually used to describe six or more missing teeth, and anodontia is the complete absence of teeth (Figures 1 and 2).

Hypodontia can also be classified, according to the severity of the condition, as:
- mild: one to three teeth developmentally missing;
- moderate: four to six teeth developmentally missing; and,
- severe: more than six teeth developmentally missing.

Hypodontia can affect both the primary and permanent dentition. It is rare in the primary dentition, with a prevalence of less than 1%
in Caucasians. When it does occur in the primary dentition, it most commonly involves the mandibular incisors.2

Aetiology
Developmental absence of teeth is a consequence of:
- physical obstruction or disruption of the dental lamina;
- space limitation;
- functional abnormalities of the dental epithelium; or,
- failure of initiation of the underlying mesenchyme.3

Hypodontia may arise as a familial condition, with a high proportion of affected individuals coming from families with a previous history of the condition.4 The nature of the inheritance is complex and not well understood, but it is thought to be related to more than one gene.5,6 Hypodontia may also arise in individuals with no family history. A number of systemic conditions, such as hypohidrotic ectodermal dysplasia, Down syndrome and chondroectodermal dysplasia have hypodontia as a feature. The developmental disruption due to the presence of a cleft lip and palate involving the alveolus may also result in an absence of teeth in that region, notably the maxillary lateral incisors.7,8

Teeth develop as appendages of the embryonic surface epithelium. The most important events during regulation of the development of all such organs are the so-called inductive interactions.6 Signal molecules of several different families are used sequentially during the advancing development, and reciprocally from epithelium to mesenchyme and vice versa. Signalling interactions that determine the location, identity, size, and shape of teeth take place during the early stages of tooth development. The first signals are secreted by the oral ectoderm, which initiates the odontogenic programme in the underlying neural crest-derived mesenchyme. The committed mesenchyme signals back to the epithelium and controls the growth and folding of the epithelium. The mesenchymal signals also induce the formation of signalling centres in the epithelium, in which many genes encoding signal molecules are activated. These centres signal back to mesenchyme, as well as within the epithelium, and regulate the advancing development, including cusp development in molars. Numerous transcription factors have been identified, which are turned on in the target tissues as a result of signalling.

Prevalence
Studies assessing hypodontia vary widely in their reports of prevalence in the permanent dentition, as can be seen in Table 1. The majority of these studies report prevalence rates varying from 2.6% in Saudi Arabia9 to 11.3% in Ireland.10 Studies in the United Kingdom suggest a prevalence rate of 4.4.5%.11,12 Some of these studies are biased because of the nature of the population studied, i.e., orthodontic patients, and hence these figures cannot be generalised for the whole population.

Management
The developmental absence of teeth can seriously disable a young person, both physically and emotionally, especially during the turbulent years of adolescence. Early management is indicated, yet there are reports of patients with hypodontia being referred late with all that that implies – treatment disrupting the examination years and reluctance to wear appliances because of the impact on the young person’s social life.13

There is much to be gained from the interdisciplinary management of young people who have hypodontia. Many patients are looked after by multidisciplinary teams, and each clinician does their own treatment in isolation. True interdisciplinary working involves the close working of a committed team where each member contributes their expertise to achieve an optimum outcome for the patient and their family.3,13,14
Background
Prior to 2001 the treatment of patients with developmental dental disorders in Ireland depended on the region where the patient was resident and was not co-ordinated. In December 2001 a Special Dental Needs Restorative Dentistry Clinic was established and funded by the Department of Health and Children to treat this patient population.

The purpose of this study is to identify the prevalence, frequency, severity, and geographic distribution of hypodontia cases referred to a tertiary care centre for developmental dental disorders, where an interdisciplinary approach for the management of such cases is currently available.

Materials and methods
The details of 168 patients with hypodontia referred to the Special Dental Needs Clinic in the Dublin Dental School and Hospital were entered into an Access Microsoft database designed as a national record for patients with developmental dental disorders, which included hypodontia, amelogenesis imperfecta, and dentinogenesis imperfecta. Ethical approval was obtained from the Faculty Research Committee in Trinity College Dublin and informed consent obtained from each patient or parent. Teeth charting was completed using both clinical examination and orthopantomographs. All data were entered by one investigator.

The Special Dental Needs Clinic is specifically for the treatment of patients with developmental craniofacial and dental anomalies. The main categories include:

1. Dental anomalies:
- moderate and severe developmental hypodontia;
- amelogenesis imperfecta;
- dentinogenesis imperfecta;
- microdontia/macrodontia (anomalies of tooth structure, size or eruption); and,
- failures of eruption.

2. Developmental disorders with associated craniofacial/dental anomalies:
- osteogenesis imperfecta;
- epidermolysis bullosa – recessive dystrophic type;
- ectodermal dysplasias; and,
- cleidocranial dysplasia.

Results
Referred cases
Of all patients referred to the clinic, hypodontia cases constituted 65.5% of referrals, amelogenesis imperfecta cases represented 28.5%, and dentinogenesis imperfecta cases represented 6% of the total referrals. This paper will only describe the hypodontia population.

Sex distribution
Females were more affected than males with a ratio of approximately 1.3:1.

Source of referrals
Most referrals were from the Dublin North East and Dublin Mid-Leinster areas, with fewer referrals coming from the west and south of the country (Figure 3). The majority of referrals were from the Health Service Executive (previously known as the Health Board Dental Service), followed by referrals from general dental practitioners (Figure 4). Very few cases had been self-referred or referred by a general medical practitioner.

Table 1: Previous hypodontia prevalence studies.1

<table>
<thead>
<tr>
<th>Country</th>
<th>Author</th>
<th>Population type</th>
<th>Age range (yr)</th>
<th>Number of patients</th>
<th>Prevalence (%)</th>
<th>Most frequently absent tooth</th>
</tr>
</thead>
<tbody>
<tr>
<td>Malaysia</td>
<td>Nik-Hussein, 1989</td>
<td>Children attending</td>
<td>6-15</td>
<td>1,583</td>
<td>2.8%</td>
<td>Maxillary lateral Incisor</td>
</tr>
<tr>
<td>Saudi Arabia</td>
<td>Salama and Abdel-Megid, 1994</td>
<td>Children attending</td>
<td>5-10</td>
<td>1,300</td>
<td>2.6%</td>
<td>Mandibular second premolar</td>
</tr>
<tr>
<td>Australia</td>
<td>Lynham, 1990</td>
<td>Australian defence</td>
<td>16-26</td>
<td>662</td>
<td>6.3%</td>
<td>Maxillary lateral incisor</td>
</tr>
<tr>
<td>Norway</td>
<td>Aasheim and Ogaard, 1993</td>
<td>Schoolchildren</td>
<td>7.8-10.4</td>
<td>1,953</td>
<td>6.5%</td>
<td>Mandibular second premolar</td>
</tr>
<tr>
<td>Iceland</td>
<td>Johannsdottir et al, 1997</td>
<td>Schoolchildren</td>
<td>6</td>
<td>396</td>
<td>5%</td>
<td>Mandibular second premolar</td>
</tr>
<tr>
<td>Denmark</td>
<td>Rolling, 1980</td>
<td>Schoolchildren</td>
<td>9-10</td>
<td>3,325</td>
<td>7.8%</td>
<td>Maxillary second premolar</td>
</tr>
<tr>
<td>Hong Kong</td>
<td>Davis, 1987</td>
<td>Schoolchildren</td>
<td>12</td>
<td>1,093</td>
<td>6.9%</td>
<td>Mandibular incisor</td>
</tr>
<tr>
<td>Ireland</td>
<td>O’Dowling and McNamara, 1990</td>
<td>Orthodontic patients</td>
<td>7-17</td>
<td>3,056</td>
<td>11.3%</td>
<td>Mandibular second premolar</td>
</tr>
<tr>
<td>England</td>
<td>Rose, 1966</td>
<td>Orthodontic patients</td>
<td>7-14</td>
<td>6,000</td>
<td>4.3%</td>
<td>Mandibular second premolar</td>
</tr>
<tr>
<td>England</td>
<td>Brook, 1974</td>
<td>Nursery and schoolchildren</td>
<td>3-5 &amp; 11-14</td>
<td>958 &amp; 1,183</td>
<td>4.4%</td>
<td>Mandibular second premolar</td>
</tr>
<tr>
<td>United States</td>
<td>Muller et al, 1970</td>
<td>School students</td>
<td>11-15</td>
<td>14,940</td>
<td>3.5%</td>
<td>Mandibular second premolar</td>
</tr>
<tr>
<td>Sweden</td>
<td>Bergstrom, 1977</td>
<td>Schoolchildren</td>
<td>8-9</td>
<td>2,589</td>
<td>7.4%</td>
<td>Mandibular second premolar</td>
</tr>
</tbody>
</table>
Pattern of tooth agenesis

The mandibular second premolars showed the highest frequency of tooth agenesis, representing 15% of the total number of missing teeth, followed by the maxillary second premolars (14.4%) and the lateral incisors (13%). The distribution of tooth agenesis is shown in Figure 5. The total number of missing teeth for all patients was 1,431, with a range of two to 26. Symmetry of tooth agenesis of the right and left sides was a feature in individual patients; however, more teeth were missing on the left side (n = 725) as compared to the right side (n = 706), and in the maxillary arch (n = 768) as compared to the mandibular arch (n = 663). The most common pattern of tooth agenesis per patient was four missing teeth, followed by two missing teeth per patient. A total of 91 patients (54%) had severe hypodontia, where more than six teeth were developmentally missing, and 54 patients (32%) had moderate hypodontia, where four to six teeth were developmentally missing.

Discussion

Hypodontia was the most common developmental dental disorder referred to the Special Dental Needs Clinic since its inception in 2001. By definition, this patient population is biased and does not reflect the prevalence and severity of hypodontia for the Irish population. The results should be interpreted bearing this limitation in mind.

In agreement with other studies, females were more commonly affected than males; this may suggest a referral bias, as females are more likely to seek dental treatment than their male counterparts.

The majority of referrals came from the Health Service Executive and were from in and around the Dublin area. This may reflect the density of the population in this area as 50.6% of the population lives in the Dublin Mid-Leinster and Dublin North East areas, whereas 25.5% lives in the south and 24% lives in the west of the country. The age range was seven to 50 years, with a median age of 20 years (mean: 21.79; SD: 8.005). To exclude other causes of tooth loss for older patients, clinical notes and referral letters were checked carefully, and compared with previous radiographs available in the charts. While recognition of the younger patients with hypodontia results either from chance observation or a positive family history, it might be expected that the majority of cases would be identified in the mixed dentition phase. The median age of this patient population is 20 years; this may suggest either poor recognition by dental practitioners, or low levels of patient demand due to the prohibitively expensive cost of treatment. For these cases, it is likely that there were difficulties in locating an appropriate clinic to which a referral for advice or treatment would be made. A second possible reason is that the database was established by the Division of Restorative Dentistry and Periodontology, which traditionally provides restorative care at the end of the treatment plan, as compared to the interdisciplinary approach that has been implemented recently. Patients are now being added to the database as soon as their diagnosis is confirmed, often on referral from paediatric dentistry, community and orthodontic clinics, and a treatment plan drawn up in interdisciplinary clinics. The frequency and distribution of tooth agenesis (as shown in Figure 5) is similar to that reported by other workers, however, a slight increase in the prevalence of missing maxillary second premolars as compared to maxillary lateral incisors was noticed. This may be attributed to the fact that the majority of these patients are referred because of moderate to severe hypodontia and that mild hypodontia cases, where there were no additional features like microdontia, impactions, or failure of eruption, were not referred but treated locally. Our results were also in agreement with the results of a Swedish study. However, that group reported an increased prevalence of mandibular central incisor aplasia and did not report any agenesis of maxillary central incisors.

The most frequently reported number of missing teeth was four missing teeth per patient, followed by two missing teeth per patient (as shown in Figure 6).

The Special Dental Needs Clinic is a specialist clinic and hence most of the cases referred had moderate to severe hypodontia with four or more teeth missing (Figure 7); some mild cases were also treated when additional features such as microdontia, impacted teeth, or primary failure of eruption, which may complicate the management of such mild cases, co-existed.
Conclusion
This paper describes the profile of patients with hypodontia attending a tertiary care clinic for developmental dental disorders in the Dublin Dental School & Hospital. Hypodontia is the most common developmental disorder affecting teeth in this referred population. The range of missing teeth was two to 26, and the mandibular second premolar was the most frequently absent tooth type.

References