

Pattern of permanent teeth present in individuals with ectodermal dysplasia and severe hypodontia suggests treatment with dental implants

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Abstract

Purpose: *The objective of this study was to assess the pattern of permanent teeth present in a self-selected sample of 17 female and 35 male patients with ectodermal dysplasia presenting for treatment with dental implants.*

Methods: *Mean age of sample: 18.7 years, age range: 5.9 to 60.9 years. Panoramic radiographs were examined independently by two investigators to determine the permanent teeth present. None of the sample reported extractions of permanent teeth prior to presenting for implants.*

Results: *Permanent teeth most likely to be present, reported as a percentage of the patient sample with that tooth, were: maxillary central incisors (42%), maxillary first molars (41%), mandibular first molars (39%), maxillary canines (22%), mandibular second molars (17%), maxillary second premolars (15%), and mandibular premolars (12%). Comparing dentition by quadrants, mandibular anterior teeth (canines and incisors) were least likely to be present.*

Conclusions: *Results support previous findings that the maxillary central incisors, maxillary first molars, mandibular first molars, and maxillary canines are the most conserved teeth in hypodontia. Successful use of osseointegrated implants in the anterior mandibles of most of these patients suggests that habilitation of the mandible with dental implant-supported prostheses is a reasonable option. (Pediatr Dent 20:278-80, 1998)*

Ectodermal dysplasia (ED) is an inherited disorder in which at least two structures derived from the ectoderm are abnormal. ED may be inherited by all Mendelian means of inheritance.¹ Signs and symptoms have been well described.^{2,3} Oral findings can be significant and include multiple tooth abnormalities including anodontia and hypodontia with associated lack of normal alveolar ridge development.⁴ Other physical signs may involve the sweat glands, scalp hair, nails, skin pigmentation, and abnormal or underdevelopment of craniofacial structures.^{5,6}

The condition includes two major types, hypohidrotic, in which the sweat glands are absent or decreased significantly, and hidrotic, in which the sweat glands are normal. The hypohidrotic is the more severe type and is associated with sensitivity to heat and frequent high fevers. This type is also thought to have more associated dental defects. Hypohidrotic ED is an X-linked recessive trait. Birth prevalence of X-linked hypohidrotic ED has been estimated to be between 1/10 000 and 1/100 000.⁷ The gene responsible for hypohidrotic ED has recently been identified.⁸

Hypodontia is the second most frequently reported sign in ED (80% of cases).¹ The pattern of missing teeth in severe hypodontia has been reported in previous studies.⁹⁻¹¹ Although Schalk-van der Weide and colleagues found no association between congenitally missing teeth and ectodermal symptoms in a study comparing dental findings in 167 patients with oligodontia with 135 normal controls, they suggested that an individual with the most stable teeth missing, or with several teeth missing, should be examined carefully for signs of ED.⁹ In another study, individuals with hypodontia associated with a syndrome were found to be more likely to be missing more teeth than individuals with hypodontia not associated with a syndrome.¹⁰ In a sample of patients with X-linked hypohidrotic ED, Crawford found more teeth missing in males and a difference in the pattern of teeth missing between males and females.¹¹ Thesleff has recently summarized information concerning the genetic control of tooth development.¹²

Additional information concerning the pattern of permanent teeth present in severe hypodontia may contribute to knowledge concerning etiological factors in tooth agenesis.¹³ Also, this information may be helpful in formulating a general approach to planning prosthodontic treatment of individuals with ED and severe hypodontia. The purpose of this study was to assess the pattern of permanent teeth present in a self-selected sample of patients with ED presenting for treatment with dental implants.

Methods

The self-selected patient sample consisted of 17 females and 35 males diagnosed as having a form of ED with associated severe hypodontia, who presented to the dental clinic at the National Institute of Dental Research, National Institutes of Health, for treatment with dental implants. The participants were responding to information concerning a dental implant clinical trial distributed by the National Foundation for Ectodermal Dysplasia (National Foundation for Ectodermal Dysplasias, 219 East Main, PO Box 114, Mascoutah, IL 62258-0114). Mean age of the sample was 18.7 years, with an age range from 5.9 to 60.9 years.

Panoramic radiographs of the sample were obtained and examined independently by two investigators using a standard radiographic viewing box to determine the permanent teeth present. None of the participants reported extractions of permanent teeth prior to presenting for treatment with dental implants. Disagreements between investigators concerning tooth identification were resolved by consensus.

Results

The permanent teeth most likely to be present reported as a percentage of the patient sample with that tooth present were: maxillary central incisors, present in 42% of the sample, maxillary first molars (41%); mandibular first molars (39%); maxillary canines (22%); mandibular second molars (17%); maxillary second premolars (15%); and mandibular premolars (12%) (Fig 1).

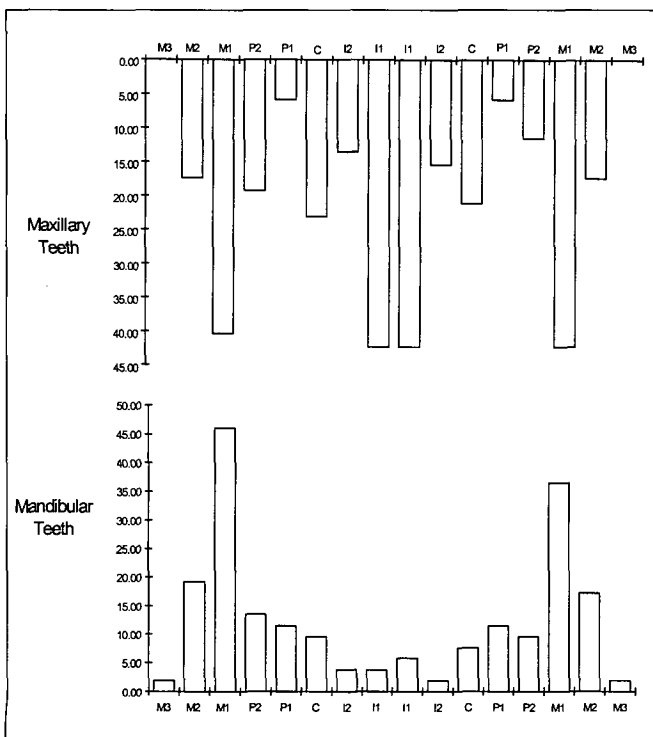


Fig 1. Percent of patients with each permanent tooth present.

Comparing dentition quadrants, mandibular anterior teeth (canines and incisors) were least likely to be present (Wilcoxon's signed-rank test, $P < 0.01$). No significant differences were found between males and females.

Discussion

The results support those of other studies in that the maxillary central incisors, maxillary first molars, mandibular first molars, and maxillary canines are the most conserved teeth in severe hypodontia.^{10, 14, 15}

Because males with ED usually exhibit more signs than females, a random sample of ED patients with hypodontia would be expected to demonstrate more missing teeth in males than females.¹¹ However, a prerequisite for participation in the protocol was severe hypodontia associated with a form of ED. Consequently, all patients had severe hypodontia and were seeking treatment with dental implants. This likely obscured gender differences in severity of hypodontia.

Considerable research supports the efficacy of rehabilitating a completely or partially edentulous mandible using a prosthesis supported by implants anchored in the anterior mandible.^{16, 17} The congenital absence of teeth can result in minimal bone support for complete or partial, removable prostheses.

The congenital absence of teeth can result in minimal volume of bone for the placement of endosseous implants in locations favorable for subsequent restorations. However, most of the patients whose radiographs comprised this sample, were successfully treated with mandibular fixed/detachable or bar-clip prostheses supported by implants.¹⁸ Preliminary results from the implant phase of this study, which included most of this sample, found that 203 of 243 cylindrical threaded implants (92%) placed primarily in the anterior mandibles of 52 patients aged 7 to 68 years with severe hypodontia successfully osseointegrated and were still functioning after 3 years. Success rates varied by age group, 87% in preadolescents (aged 7–11), 90% in adolescents (aged 12–17), and 97% in adults (older than 17). We have reported previously on a child who had implants placed in the anterior mandible at age 3.5 years that successfully osseointegrated and functioned to support a bar-clip overdenture after 5 years.¹⁹

Benefits of placing implants in young patients have not been demonstrated. Bone volume may not be sufficient for placing the implants in ideal positions to support the prosthesis. Also, craniofacial growth will necessitate remake and redesign of the prosthesis as growth occurs. If implant-supported prostheses were shown to have positive effects on craniofacial growth, social development, self-image, and food choice, their use in the anterior mandible might be routinely recommended in younger patients.

Summary

The results of this study support previous findings that the maxillary central incisors, maxillary first molars, mandibular first molars, and maxillary canines are the most conserved teeth in severe hypodontia. Because of the frequency of missing permanent mandibular anterior teeth, individuals with ED and severe hypodontia may be candidates for prosthodontic management with endosseous implant-supported mandibular dentures.

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