



Use of endosseous implants in a 3-year-old child with ectodermal dysplasia: case report and 5-year follow-up

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The ectodermal dysplasias represent a group of inherited disorders characterized by defects in tissues that are derived from ectoderm. Freire-Maia and Pinheiro describe more than 100 different taxonomic groupings of ectodermal dysplasia across a wide spectrum of clinical presentation.¹

The classic form of ectodermal dysplasia (Christ-Siemens-Touraine syndrome) is thought to be X-linked and involves hypodontia, hypohidrosis, hypotrichosis, and a characteristic facies. Because these individuals don't sweat, this condition is sometimes referred to as X-linked, hypohidrotic ectodermal dysplasia (XLHED).

The dental literature describes many conventional prosthetic approaches to the clinical management of these patients.²⁻⁶ Osseointegration is well documented as a safe and predictable method of tooth replacement.^{7, 8, 9} The early clinical research was done in edentulous adults and subsequent studies have confirmed the successful application of this modality in a variety of clinical situations.^{10, 11} Almost all of this scientific investigation, however, has been performed in adults, when the dynamics of growth and development are not an issue.

Osseointegrated implants are in direct apposition to bone and lack the compensatory mechanism of a periodontal ligament. Research models indicate that osseointegrated fixtures do not move with the growth of the jaws and suggest caution in their use in individuals where growth is incomplete.^{12, 13}

However, the lack of relevant long-term clinical studies has not prevented clinicians from using implant-assisted prostheses in children. The literature contains several anecdotal reports of the use of dental implants in children, many with anodontia or severe hypodontia, often associated with ectodermal dysplasia, or from trauma.¹⁴⁻²¹

The purpose of this paper is to present a case report of implant placement in a patient 3 years and 3 months of age, subsequent prosthodontic treatment and 5-year follow-up. We believe this to be the youngest patient documented to have been treated with a dental implant supported dental prosthesis.

Case report

History

A 3-year-old white male diagnosed with Christ-Siemens-Touraine ectodermal dysplasia was referred to the NIDR dental clinic for prosthetic evaluation following implant placement (Fig 1). The implants had been placed by an oral and maxillofacial surgeon in private practice. The patient's mother reported that the rationale for and possible complications of implant placement in a 3-year-old child were not discussed with her prior to placing the implants.

The patient was referred to the National Institute of Dental Research (NIDR) by the National Foundation for Ectodermal Dysplasia* for possible participation in an ongoing protocol.

The patient was 3 years and 7 months old at presentation and had four mandibular and two maxillary IMZ press fit (Interpore International, Irvine, CA), endosseous implants, which had been placed at age 3 years 3 months. Hydroxyapatite of unknown type was also placed to augment both the maxilla and mandible and is readily seen on radiographs (Fig 2). This patient had only two tooth buds, both located in the maxilla, and had never worn a dental prosthesis.

Since the patient had already received implants prior to presenting at NIDR, he was not eligible for the implant protocol. However, be-



Fig 1. Patient with X-linked hypohidrotic ectodermal dysplasia and severe hypodontia at 3 years 7 months. Endosseous dental implants were placed 4 months earlier.

* National Foundation for Ectodermal Dysplasia, 219 East Main, PO Box 114, Mascoutah, IL 62258.

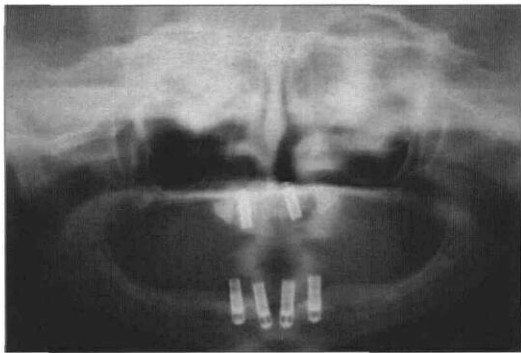


Fig 2. Panoramic radiograph of patient taken at time of clinical exam. Two maxillary canine tooth buds and 6 IMZ implants are evident. HA is also seen, especially in the mandible.

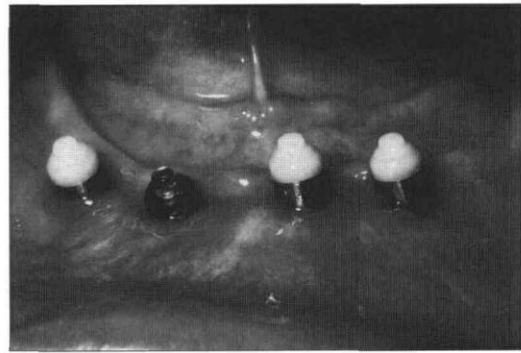


Fig 3. One month following abutment connection. Soft tissue healing was uneventful.

cause of our interest in the use of oral implants in children, the patient was accepted for prosthodontic treatment and follow-up care under an NIDR omnibus diagnosis and treatment protocol.

Clinical exam

The clinical findings were consistent with the diagnosis of X-linked hypohidrotic ectodermal dysplasia. No teeth were clinically present, and the implant surgical sites had healed uneventfully. The intraoral soft tissues were within normal limits, and the patient was appointed for uncovering of the implants (second-stage) surgery.

Second-stage surgery

Second-stage surgery was performed in the operating room 1 month after the initial visit. The maxillary right implant which was mobile and surrounded by hyperplastic tissue, was removed. The remaining five implants were judged to be osseointegrated (clinically immobile) and titanium conversion abutments (Implamed, Sunrise, Fl) of appropriate lengths were connected to the four mandibular implants. Since the remaining maxillary implant could not contribute to support of a prosthesis, primary closure was obtained without placing an abutment. The hydroxyapatite appeared to be stable. The gingival tissue was trimmed and approximated around the mandibular implant abutments and a final impression taken. Healing caps were placed and the patient was discharged the following day. Postoperative healing was uneventful. (Fig 3).

Prosthetic treatment

The final prosthesis was completed within 2 months of the second-stage surgery. This consisted of a conventional maxillary denture and a mandibular overdenture supported by two cast gold bars, which were secured to the implants and separated in the midline. During the 5-year follow-up period the prosthesis was remade or relined as appropriate to accommodate eruption of the maxillary teeth and overall facial growth (Fig 4).

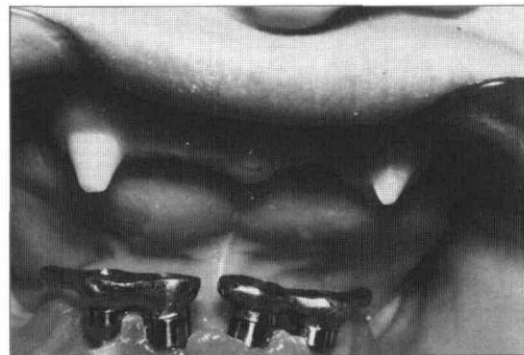


Fig 4. 14 months after prostheses completion. All implants are stable and the soft tissue is within normal limits. The prostheses were remade at this point to accommodate the eruption of the two maxillary teeth and overall growth. The patient is aged 4 years and 11 months.

Cephalometric and panoramic radiographs were taken 5 years after initial implant placement to assess the bone height and the relative position of the implants and the hydroxyapatite (Figs 5, 6 & 7).

Follow-up

This patient has experienced significant growth since the endosseous implants were placed. The mandibular implants have been in function for 4 years. Their relative position within the anterior mandible remains unchanged as growth takes place in the rami and condyles. This is consistent with mandibular growth described by Bjork.²² The fate of the hydroxyapatite remains uncertain. After a five-year clinical follow-up, the ridges remain firm and there is no inflammation or evidence of exfoliation. However, after 5 years there is radiographic evidence that the HA is being resorbed. The impact of this resorption on the integrity of the implants in the long term is uncertain. The most dramatic finding is the relative position of the unloaded single maxillary implant fixture. It is clear that this implant has not moved with the downward and forward growth of the maxilla. It now is positioned in close proximity to the floor of the nose. With further

significant growth remaining, the ultimate position of this implant is uncertain. This implant will have to be followed closely and possibly removed surgically.

Discussion

This patient presented a significant opportunity to follow the consequences of placing of oral implants in a very young patient. We believe this may be the youngest patient to receive dental implants to support an intraoral prosthesis. The patient's mother reported that other treatment options were not discussed with her, nor was she informed of possible complications of placing implants in a very young child. In addition, the cost of the necessary prosthodontic treatment and subsequent modifications

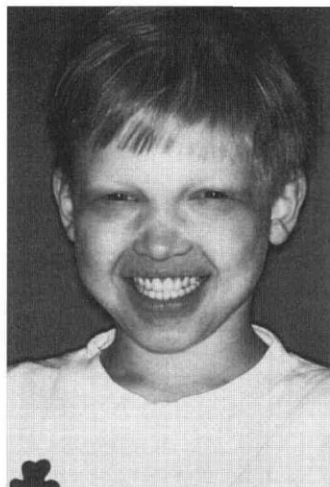


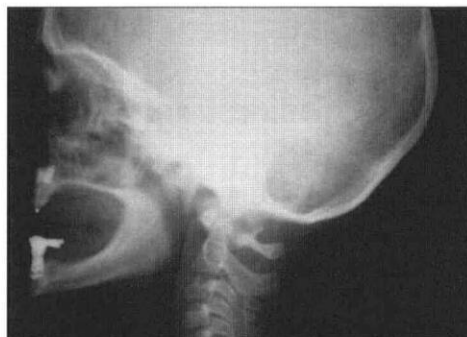
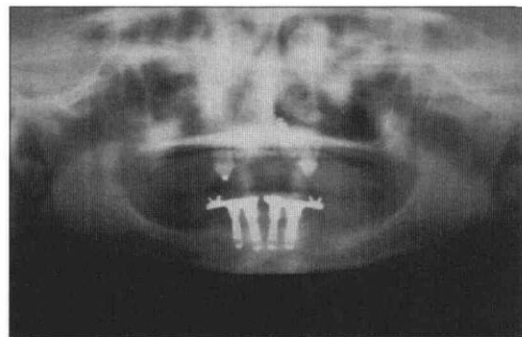
Fig 5. Patient at 8 years and 3 months of age. The maxillary and mandibular overdentures were remade. The implants were placed 5 years previously.

plants are not intermixed with natural teeth. However, the cost of maintaining and remaking the prosthesis as a young patient matures will probably be significant. While the magnitude of the change in position of the maxillary implant observed in this patient may not be typical, it provides a cautionary note regarding implant placement in the growing maxilla.

The most significant issue raised by this case is if and when treatment of a young child with implants is indicated and prudent. There is little research identifying any physiologic benefits of placing implants in children.

The theoretical possibility exists that significant edentulism in a growing child adversely affects craniofacial growth.²³⁻²⁵ However, while the literature supports the concept that craniofacial development is adversely affected in individuals with ectodermal dysplasia, the exact mechanism by which the growth is affected is unclear. The most obvious effect of ectodermal dysplasia is the lack of alveolar bone in areas of anodontia, which significantly decreases the tissue support for removable prostheses and also can make

implant placement more difficult. Limited evidence in adults shows that the increased functional load on the mandible following treatment with an implant-supported prosthesis may be associated with an increase in mass of the mandible. This



Figs 6 & 7. Panoramic and cephalometric radiographs. The unloaded maxillary implant has become deeply imbedded with the growth of the maxilla.

and remakes of the prostheses as the child matured were not discussed.

This case report confirms that implants placed in the anterior mandible will move with the mandible as growth occurs in the condyles and rami. The rotation of the mandible, which accompanies growth apparently has not caused a significant problem relative to the angulation of these implants and the prosthodontic occlusal plane. The change in position of the maxillary implant as the mandible grows downward is not unexpected. This position change is a significant complication for this patient. Eventually it may be necessary to remove this implant.

Cronin and Oesterle have discussed the possible growth-related problems of placing implants in young patients.^{18,19} They discussed the possible complications that an ankylous implant fixture in a young patient might present as growth occurred. Problems with implants placed in the mandible may be minimal if im-

evidence may provide a hypothetical reason to use implants to support a mandibular prostheses in the mandible of an edentulous child.^{26,27} However, at present there is no evidence from clinical trials to indicate that implant placement in young children has a positive effect on craniofacial growth and development.

Investigators have found a psychological benefit of placing implants in children aged 12-19.²⁸ However these data were obtained shortly after prostheses placement and could have been a result of a treatment effect. Data from well-controlled studies concerning the physiologic and psychological effects of placing implants in young children are not available.

In summary, psychological benefits may be associated with using implants to support an oral prosthesis in the mandible of a teenage child with many missing teeth. However clinical research has not demonstrated compelling reasons to place implants in preteenage children to support an oral prosthesis. Carefully con-

trolled prospective clinical studies are needed to determine the efficacy and effectiveness of the use of implants in children and young adults.

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