# Treatment outcomes for adolescent ectodermal dysplasia patients treated with dental implants

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**Summary.** To evaluate dental implant survival in patients with ectodermal dysplasia (ED). To assess patterns of hypodontia in this patient group.

*Methods*. A retrospective analysis of the use of dental implants in ED patients treated at the Royal Children's Hospital, Melbourne.

*Results.* Sixty-one implants were placed into 14 patients (nine male and five female). The mean age of patients receiving maxillary implants was 18 years 6 months (range 17 years 9 months–20 years 0 months) and mandibular implants was 17 years 5 months (range 12 years 2 months–21 years 11 months). The mean follow-up period was 3 years 4 months (range 1 year 18 months–5 years 1 month). Forty-three implants were placed in the anterior mandible, three in the posterior mandible and the remaining 15 in the anterior maxilla.

Of the 61 implants placed, 54 [88.5%] successfully integrated and were able to be restored. Three of the 15 implants placed into the anterior maxilla [20%] failed, while four of the 46 in the anterior mandible failed [8.7%]. Five of the 14 patients [35.7%] had at least one implant fail prior to abutment connection.

At the 12-month review appointments, 41 of the integrated 54 implants [76%] were reviewed and classed as successful, giving an overall success at follow up of 67.2%. Thirteen implants [21.3%] were unable to be reviewed owing to geographical reasons.

Teeth most likely to be present in the maxilla were the central incisors [71%], first molars [54%] and canines [43%], whereas in the mandible they were the canines [53%] and the first premolars and first molars [40%].

*Conclusions.* Dental implants can be placed, restored and loaded in ED patients. Maxillary teeth most likely to be present are the central incisors, canines and first molars, whereas in the mandible the canines, first premolars and molars are most likely to be present. Prior to cessation of growth, implant placement in the symphyseal region of the anterior mandible may be performed with caution. Despite the limited numbers and with due consideration to jaw development, the results support the continual use of endosseous dental implants in this group of patients for optimal clinical outcomes.

#### Introduction

Ectodermal dysplasia (ED) is a general term representing a group of inherited disorders characterized by aplasia or dysplasia of tissues of ectodermal origin. These include skin, hair, nails, teeth, sweat glands, nerve (neural) cells and constituent parts of the ear and eye.

Freire-Maia and Pinheiro [1] described 117 variants using multiple secondary signs unique to each syndrome to confirm the diagnosis. Clinical features include hypodontia or anodontia (missing teeth), trichodysplasia (abnormal hair), dyshidrosis (abnormal sweat glands), asteatosis (abnormal sebaceous glands)

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and onychodysplasia (abnormal nails). Common extraoral signs include sparse fine or coarse curly hair, abnormally developed nails, frontal bossing, prominent lips, a depressed midface and nasal bridge, and skin that is soft, thin and dry and often subject to eczema. Hypodontia occurs in 80% of cases.

Clinically the condition may be divided into two groups: hidrotic and anhidrotic (hypohidrotic) forms. The hidrotic form, inherited as an autosomal trait, does not usually involve sweat glands and was first described by Clouston [2] in 1929. Whereas the anhidrotic form, classified as X-linked recessive, is characterized by a triad of hypodontia, hypohidrosis and hypotrichosis and is often accompanied by characteristic facial features including a prominent forehead, wide eyebrows, saddle-shaped nose, thick everted lips, dry skin and sparse hair [3]. The resultant hypodontia results in reduced alveolar bone growth, and lack of development of the alveolar ridges, which clinically often appear to be extremely narrow and concave lingually. If teeth are present, they are often conical in shape, malformed and widely spaced [4]. The lack of teeth and subsequent inhibited development of the alveolar ridges may result in reduced vertical facial height. Recently, the gene responsible for anhidrotic ectodermal dysplasia was identified [5].

Historically, prosthetic treatment for ED patients involved removable partial dentures, removable partial or complete overlay dentures and fixed partial dentures. The advent of dental implants has provided an additional treatment modality for restoration of the dentition in this group of patients.

There are aesthetic, functional and psychological reasons that make it important to start oral rehabilitation early in life [6]. Serial sets of dentures may be required as a child matures. Lowry [7] describes the need for the appliance to restore vertical dimension and prevent the undesirable protruding lips secondary to overclosure, thereby improving the profile. Ideally, a restored dentition should be in place before the child attends primary school [8], and some authors [9] regard oral rehabilitation of a child as having a major psychological impact on the patient's self esteem and facilitating social acceptance. Early studies in children demonstrated a marked change in a personality, speech and vocabulary 8 weeks after insertion of complete dentures [10].

The use of dental implants is a well-established treatment modality in patients who have ceased growth [11–13], and have also proved effective in certain situations in growing individuals [14,15] including those with ectodermal dysplasia [16–18].

The aim of this study was to retrospectively analyze the management of hypodontia in adolescent ectodermal dysplasia patients treated at the Royal Children's Hospital, Melbourne (RCH). The success rates of the implants and the prosthetic appliances were assessed, and the distribution of teeth present in affected individuals and any relationship between the prosthodontic appliance and subsequent growth was also evaluated.

## Patients and methods

All patients referred to the Department of Dentistry at the RCH, with a diagnosis of ectodermal dysplasia and associated hypodontia, who have been treated using dental implants were included in this study. The group included 14 patients (five females and nine males), with an age range between 12 years 2 months and 21 years 11 months [see Tables 1 and 2].

Prior to treatment, each patient underwent a multidisciplinary team consultation which included a paediatric dentist, prosthodontist, periodontist, oral and maxillofacial surgeon and an orthodontist. The need for adjunctive procedures was assessed and documented and treatment plans formulated based on the needs of each individual child.

Clinical and radiographic examinations were performed, including orthopantomogram and intraoral radiographs, as required. The number of missing teeth was recorded, as were the position and number

Table 1. Demographic data of ectodermal dysplasia patients treated with implants at the Royal Children's Hospital.

	Total number of patients	n
Mean age at implant placement (mandible)	14	17 years 5 months (range: 12 years 2 months-21 years 11 months)
Mean age at implant placement (maxilla)	4	18 years 6 months (range: 17 years 9 months-20 years 0 months)
Mean age at implant restoration		19 years 3 months (range: 12 years 8 months-23 years 6 months)
Mean follow up since implant placement		3 years 4 months (range: 1 years 6 months-5 years 1 months)
Mean follow up since restoration		2 years 5 months (range: 1 years 0 months-4 years 8 months)

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Patient	Arch	Sex	Age at implant surgery
1	Mandible	М	20 years 0 month
	Maxilla		20 years 4 months
2	Mandible	F	18 years 0 month
3	Mandible	F	16 years 6 months
4	Mandible	F	19 years 8 months
5	Mandible	F	20 years 4 months
6	Mandible	Μ	18 years 9 months
7	Mandible	М	21 years 11 months
8	Maxilla	М	17 years 9 months
	Mandible		18 years 3 months
9	Mandible	М	20 years 10 months
10	Mandible	М	12 years 2 months
11	Mandible	F	19 years 7 months
12	Maxilla	М	18 years 3 months
	Mandible		18 years 3 months
13	Maxilla	М	18 years 0 month
	Mandible		18 years 0 month
14	Mandible	М	12 years 4 months

Table 2. Age at implant placement, sex and arch treated in ectodermal dysplasia patients.

of teeth present. A review of any current prostheses was undertaken, as well as an age assessment relative to the most appropriate timing for implant placement.

#### Results

Hypodontia in ED patients occurs to varying degrees. In the current study of 14 patients, three patients were missing all permanent teeth, whereas the average number of missing teeth was 22.5. Figures 1 and 2 show the percentages of missing teeth in each arch.

The percentages of teeth present in this sample of ED patients are summarized as follows:

- Maxilla: central incisors, present in 71% of the sample, canines [43%] and first molars [54%]; and
- Mandible: canines, present in 53% of the sample, first premolars [40%] and first molars [40%].

Following treatment, the individual implants were reviewed clinically and radiographically. The criteria for clinical success of implants were: being asymptomatic in function, absence of peri-implantitis, lack of bleeding on gentle probing, lack of suppuration and marginal inflammation, and lack of mobility. The study methodology did not allow for removal of the appliance to perform active testing on individual implants, as it was deemed inappropriate to subject young adults to invasive testing and psychological trauma in the absence of any clinical



**Fig. 1.** Percentage of missing maxillary teeth in ectodermal dysplasia patients treated at Royal Children's Hospital, from the maxillary right third molar (18) to the maxillary left third molar (28).



**Fig. 2.** Percentage of missing mandibular teeth in ectodermal dysplasia patients treated at Royal Children's Hospital, from the mandibular right third molar (48) to the mandibular left third molar (38).

problems. Prosthodontic success was evaluated using the criteria described by Walton [19].

Of the 61 implants placed, 54 were integrated and able to be restored, giving an integration rate of 88.5%. Fifteen maxillary implants were placed in four subjects while 46 mandibular implants were placed into 14 subjects. Mean ages at implant placement, restoration and follow up are shown in Table 1, age at placement, sex and arch treated are summarized in Table 2. In total, 14 fixed and three removable prostheses were placed, as shown in Table 3. An analysis of implant length and diameter is shown in Table 4 and Fig. 3. The lack of alveolar ridge development resulted in the need for narrow platform implants in three patients.

Seven of the 61 implants [11.4%] failed prior to abutment connection. In one patient, two implants

Table 3. Summary of implants and prosthesis placed in the study.

No. of implants placed	61
No. of implants restored	54
Integration rate	88.5%
Removable prosthesis	3
Fixed prosthesis	14

failed in the maxillary premolar region. This patient had previously undergone a maxillary osteotomy and iliac crest graft, and presented with chronic sinusitis and mobile implants, which were removed. In this case, however, a sufficient number of implants remained to allow construction of a prosthesis. In another patient an implant failed in the 35 region. The alveolus in this area was narrow and the implant was placed in close proximity to the permanent first bicuspid tooth. A third patient lost an implant in the 43 region. This implant was placed immediately following the extraction of the permanent canine. Following a healing period, another implant was placed which subsequently integrated. Two implants were lost in the anterior mandible of a patient who had previously had two horizontally impacted canines removed. Following a suitable healing period, two further implants were placed, which subsequently integrated.

Maxillary implants accounted for 15 of the 61 implants placed. Of these, 12 successfully integrated and were subsequently restored. Forty-two of 46 mandibular implants integrated and were successfully restored. Of the 14 patients, five [35.7%] had at least one failed implant prior to abutment connection. All patients were, however, ultimately provided with implant supported prosthesis.

Review appointments were arranged at 6 and 12 months post prosthesis insertion, then on an annual basis. At the 12-month review appointments, 41 of the surviving 54 implants [76%] were reviewed and classed as successful. The overall success achieved during the study at follow up was therefore 67.2%.

Four patients with 13 implants  $[21\cdot3\%]$  were unable to be reviewed owing to geographical reasons. These patients were contacted by phone and it was confirmed that no treatment or repairs had been performed. These implants were therefore classed as surviving. The remaining  $11\cdot5\%$  (seven implants) were classed as failures.

Prosthetically, 71% of all appliances were successful based upon direct clinical examination, 23% were classed as surviving based upon a telephone

**Table 4.** Summary of implant lengths and diameters. Implant lengths ranged from 10 mm to 18 mm, whereas implant diameter ranged from 3.1 mm to 4.0 mm.

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mm	10	11.5	13	15	18	Total	
3.1	2	3	2	2		9	
3.75	3	5	13	15	11	47	
4.0	3	2				5	
Total	8	10	15	17	11	61	

Summary of implants placed



Fig. 3. Number of implants placed according to length and diameter.

**Table 5.** Complications encountered during the study (n = 61 implants).

Lost implants	7
Bleeding requiring return to theatre	1
CT graft required	2
Wound dehiscence	1
Need to replace healing abutments owing to tissue irritation	1
Loose healing abutments	1
Fracture partial during healing period	4
Excessive wear with bruxing	1

assessment, whereas 6% (one patient) was classed as retreatment (repaired). All original prostheses continue to function *in situ*, with only one hybrid bridge requiring replacement of acrylic teeth owing to excessive bruxism.

Complications encountered during the study are listed in Table 5. They include loss of seven fixtures [11.4%] and a number of other complications. Beside implant loss, fracture of provisional partial dentures during the healing period was the most commonly encountered complication. In this study, no implants were placed adjacent to natural teeth in growing jaws. None of the implants placed demonstrated signs of infraocclusion during the follow-up period.

#### Discussion

When comparing the percentages of the specific teeth present there was agreement with the results of other studies [17,20,21]; however, a significantly greater number of patients in the present study had mandibular canines present [53%] compared with 10% in the study by Guckes *et al.* [17]. There was no significant difference between sexes.

Various authors have described a positive outcome with dental implants in the oral rehabilitation of young patients [6,9,10]. The hypodontia associated with ED makes these patients particularly deserving candidates for dental implant reconstruction; however, in this particular condition, the lack of bone volume in young patients, owing to failure of development of the alveolar ridges, is a major challenge in providing implant treatment. Care must be taken when assessing and treating growing patients, as the growth and development of the jaws will result in a change in implant position and angulation as the bone remodels around the 'ankylosed' implants.

The loss of implants in this study was not associated with any particular age group nor location; however, the narrow ridge anatomy often associated with ectodermal dysplasia may well be a contributing factor in the failure of some of the implants. Six of the seven failed implants were placed into sites that were deficient in bone volume or had undergone previous surgical procedures.

Guckes et al. [16] achieved 24-month survival rates of 91% in the mandible of patients with ectodermal dysplasia, whereas the success in the anterior maxilla was 71%. Statistically, they found that implants placed in the anterior maxilla were 2.8-fold more likely to fail than those placed in the anterior mandible; however, they caution over-interpretation of their results owing to the smaller number of maxillary implants placed. The same authors [17] had previously described the successful use of osseointegrated implants in the anterior mandible of ED patients. They reported 203 of 243 cylindrical threaded implants placed primarily in the anterior mandible of 52 patients aged between 7 and 68 years to have integrated and remain in function after 3 years of follow up. Their success rates were variable with age. The preadolescent group (aged 7-11) showed an 87% success rate, and adolescents [12-17] a 90% success rate, whereas the adult group showed a 97% success rate.

The youngest documented patient to receive a dental implant was 3.5 years old [18]. The child had

implants placed at 3.5 years of age in the anterior mandible, which had functioned to support a bar and clip overdenture for 5 years. It was concluded that the mandibular rotation accompanying growth had not caused a significant problem relative to the angulation of the implants and prosthodontic occlusal plane. A single implant placed in the partially edentulous maxilla of the same 3.5-year-old did, however, become deeply embedded in bone after failing to move with the subsequent dentoalveolar growth.

Smith *et al.* [22] discussed the placement of a single mandibular implant in a 5-year-old to assist in prosthodontic rehabilitation for social reasons. The patient was restored with a removable prosthesis and overdenture which was maintained over 4.5 years of function until the appliance fractured. The implant did show signs of submergence owing to dentoalveolar development associated with an erupting mandibular permanent canine, requiring the need for a longer abutment to be placed.

The use of dental implants in preadolescent patients must be carefully considered. Implants placed into developing alveolar ridges have been shown to inhibit ridge formation. All maxillary implants in this study were placed into patients above the age of 17 years who were deemed to have ceased growth. Implants placed into the anterior mandible of younger patients were used to support overdentures using ball attachments. Growth studies show little positional change of the anterior mandible as growth occurs in the rami and condyles [23].

The decision to commence implant therapy early in a child's life is a complex decision. Both the financial and biological costs need to be evaluated.

What this paper adds

<sup>•</sup> This paper describes use of dental implants in a series of 14 patients with ectodermal dysplasia.

<sup>•</sup> Patients were between the ages of 12 and 18 when implants were placed but in no case was an implant placed beside natural teeth in growing jaws.

<sup>•</sup> Of those that failed a high proportion were in areas with poor bone volume or had previously undergone surgical procedures.

Why this paper is important for paediatric dentists

<sup>•</sup> Evidence provided here shows that implants have potential to be part of successful definitive restorative treatment for some patients with ectodermal dysplasia once bone growth is complete.

<sup>•</sup> Adequate alveolar ridge anatomy may be an important factor in success.

Any perceived disadvantage of changing the abutment length or angulation, necessitating a prosthetic remake, must be weighed up against the psychological benefits of the patient receiving a more stable prosthesis [22,23].

## Conclusions

Within the limitations of the study, the following points may be concluded.

- Dental implants may be placed, restored and loaded in ED patients.
- Lack of development of alveolar ridge anatomy may be a factor associated with implant failure in ED patients.
- Maxillary teeth most likely to be present in this group of ED patients are the central incisors, canines and maxillary first molars.
- Mandibular teeth most likely to be present in this group of ED patients are the canines, first premolars and first molars.

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**Résumé.** *Propos.* 1. Evaluer la survie des implants chez les patients avec dysplasie ectodermique (ED).

2. Evaluer les patterns d'agénésie chez ce groupe de patients.

*Méthode.* Analyse rétrospective de l'utilisation des implants chez les patients atteints de ED traits au Royal Children's Hospital, Melbourne.

*Résultats*. Soixante et un implants ont été placés chez 14 patients (9 garçons et 5 filles). L'âge moyen des patients recevant des implants maxillaires était de 18 ans 6m (allant de 17ans 9m à 20 ans 0m) et des implants mandibulaires était de 17 ans 5m (allant de 12 ans 2m à 21 ans 11m). La période moyenne de suivi était de 3 ans 4m (allant de 1ans 18m à 5ans 1m). Quarante trois implants ont été placés dans la partie antérieure de la mandibule, trois dans la zone postérieure mandibulaire antérieure.

Sur les 61 implants placés, 54 (88,5%) se sont intégrés avec succès et ont pu supporter une restauration. Trois des 15 implants antérieurs maxillaires (20%) et 4 des 46 antérieurs mandibulaires ont été des échecs (8,7%). Cinq des 14 patients (35,7%) ont eu au moins un échec d'implants avant mise en charge. Lors du rappel à 12 mois, 41 des 54 implants intégrés (76%) ont été classés comme succès. Treize implants (21,3%) n'ont pu être vus pour des raisons géographiques. Les dents les plus susceptibles d'être présentes au maxillaire étaient les incisives centrales (71%), les premières molaires (54%) et les canines (43%), tandis qu'il s'agissait de canines à la mandibules (53%), des premières prémolaires et premières molaires (40%).

*Conclusions.* Les implants dentaires peuvent être places, restaurés et mis en charge chez les patients ED. Les dents maxillaires les plus susceptibles d'être présentes sont les incisives centrales, les canines et les premières molaires, tandis qu'à la mandibule ce sont les canines, les premières prémolaires et premières molaires. Avant la fin de la croissance, la mise en place d'implants dans la région symphysaire de la mandibule peut être effectuée avec précaution. En dépit du nombre limité et en prenant en compte le développement de la mâchoire, les résultats supportent l'utilisation d'implants dentaires endoosseux dans ce groupe de patients pour des résultats cliniques optimaux.

**Zusammenfassung.** *Ziele.* 1. Ermitteln der Überlebenszeit von Zahnimplantaten bei Patienten mit Ektodermaler Dysplasie (ED).

2. Bestimmen von Mustern der Hypodontie in dieser Patientengruppe.

*Methoden.* Retrospektive Studie der mit einem Implantat versorgten ED-Patienten des Royal Children's Hospital Melbourne.

*Ergebnisse*. Einundsechzig Implantate wurden bei 14 Patienten gesetzt (9 männlich, 5 weiblich). Das mittlere Alter von Patienten bei Implantation lag bei 18 Jahren 6 Monaten (Spannweite 17 Jahre 9 Monate bis 20 Jahre 0 Monate). Die mittlerer Nachbeobachtungszeit lag bei 3 Jahren und 4 Monaten (Spannweite 1 Jahr 18 Monate bis 5 Jahre 1 Monat). Vierunddreißig Implantate waren Unterkiefer-Frontbereich gesetzt worden, drei im Unterkiefer Seitenzahnbereich, die verbleibenden fünfzehn im Oberkiefer-Frontbereich.

Von den 61 Implantaten wurden 54 (88.5%) erfolgreich integriert und konnten weiter versorgt

werden. Drei der 15 Implantate im Oberkiefer-Frontbereich (20%) bzw. vier der 46 im UK-Frontbereich (8.7%) schlugen fehl. Fünf der 14 Patienten (35.7%) zeigten mindestens einen Implantatverlust vor Weiterversorgung.

Nach zwölf Monaten wurden 41 der integrierten 54 Implantate (76%) nachuntersucht und als erfolgreich eingestuft, entsprechend einer Erfolgsrate von 67.2%. Dreizehn Implantate konnten aus geographischen Gründen nicht beurteilt werden. Am wahrscheinlichsten vorhanden im Oberkiefer waren mittlere Inzisivi (71%), erste Molaren (54%) und Eckzähne (43%), während im Unterkiefer die Eckzähne am häufigsten vorhanden waren vor ersten Prämolaren und Molaren (40%).

Schlussfolgerungen. Zahnimplantate können bei Patienten mit ED gesetzt und in die Versorgung integriert werden. Im Oberkiefer waren am wahrscheinlichsten mittlere Schneidezähne, Eckzähne und erste Molaren, im Unterkiefer waren es Eckzähne, erste Prämolaren und erste Molaren. Vor Abschluss des Wachstumsalters sollte die Implantation im Bereich der Symphyse des Unterkiefers mit Vorsicht erfolgen. Trotz geringer Fallzahl und der Erfordernis der Beachtung der Kieferentwicklung legen die vorliegenden Ergebnisse eine Verwendung von enossalen Implantaten bei dieser Patientengruppe zur Verbesserung der Behandlungsergebnisse nahe.

**Resumen.** *Objetivos.* 1. Evaluar la supervivencia del implante dental en pacientes con Displasia Ectodérmica (DE).

2. Valorar los patrones de hipodoncia en este grupo de pacientes.

*Método.* Un análisis retrospectivo del uso de implantes dentales en pacientes de Displasia Ectodérmica tratados en el Hospital Real de Niños, Melbourne.

*Resultados*. Se colocaron 61 implantes en 14 pacientes (9 varones y 5 mujeres). La edad media de los pacientes receptores de implantes maxilares fue de 18a 6m (rango 17a 9m - 20a 0m) y de implantes mandibulares fue de 17a 5m (Rango 12a 2m - 21a 11m). La media del periodo de seguimiento fue de 3a 4m (Rango 1a 18m - 5a 1m). Cuarenta y tres implantes se colocaron en la mandíbula anterior, tres en la mandíbula posterior y los restantes quince en el maxilar anterior.

De los 61 implantes colocados, 54 (88,5%) se integraron con éxito y se pudieron restaurar. Fallaron tres de los 15 implantes colocados en el maxilar anterior (20%), mientras que fallaron 4 de los 46 en la mandíbula anterior (8,7%). Cinco de los catorce pacientes (35,7%) tenía al menos un implante fallido antes de la conexión del pilar.

En las citas de revisión a los 12 meses, 41 de los 54 implantes integrados (76%) fueron revisados y clasificados como exitosos, dando un éxito global en el seguimiento del 67,2%. Trece implantes (21,3%), fueron imposibles de ver debido a razones geográficas. Los dientes con más probabilidad de estar presentes en el maxilar superior fueron los incisivos centrales (71%), primeros molares (54%) y caninos (43%); mientras que en la mandíbula fueron los caninos (53%) y los primeros premolares y primeros molares (40%).

*Conclusiones*. Los implantes dentales se pueden colocar, restaurar y cargar en pacientes con DE. Los dientes del maxilar superior que con más probabilidad están presentes son los incisivos centrales, los caninos y primeros molares; mientras que en la mandíbula los caninos, primeros premolares y molares son los que con más probabilidad están presentes. Antes del cese del crecimiento, la colocación de un implante en la región de la sínfisis de la mandíbula anterior debe realizarse con precaución. A pesar del número limitado y teniendo en cuenta el desarrollo de la mandíbula, los resultados apoyan el uso continuado de implantes dentales endoóseos en este grupo de pacientes para obtener resultados clínicos óptimos.

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