

## Systematic Review Dental Implants

# Clinical outcomes of implant therapy in ectodermal dysplasia patients: a systematic review

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**Abstract.** The purpose of this review was to determine the outcome of oral function reconstruction in ectodermal dysplasia (ED) patients who have received dental implant therapy. A search was made of the PubMed and Web of Science databases; key words used were “(ectodermal dysplasia) AND (implant OR implants)”, with supplementary retrieval key words “dental implant”, “zygomatic implant”, “anodontia”, and “edentulous”. Patient age, use of bone graft, implant site, type of implant, and survival rate of the implants were included in the subsequent data analysis. Forty-five articles published between 1988 and October 2015 were included in this analysis. The cases of a total of 96 patients were retrieved (22 children and 74 adults); these patients received a total of 701 implants. Fourteen implants were removed during a median follow-up time of 24 months. The 24-month implant survival rate was 97.9% in adult subjects and 98.6% in children. Sixty-eight percent of adult patients underwent bone augmentation prior to implant placement. Based on this review, dental implants are commonly used in the oral reconstruction of ED patients. However, long-term data on bone augmentation and implant success are needed, as well as additional clinical evidence on bone resorption, the esthetic outcomes of implant therapy, and physiological considerations in ED patients.

Key words: ectodermal dysplasia; dental implant; oral function reconstruction.

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Ectodermal dysplasia (ED) is a hereditary disorder, characterized by morphological and functional hypoplasia of ectodermally derived organs and tissues. It is a non-progressive disorder. Two of the most common types of ED are X-linked recessive hypohidrotic ED (Christ–Siemens–Touraine syndrome, OMIM#305100, HED1 or HXED) and autosomal dominant hidrotic ED (Clouston syndrome, OMIM#129500,

ECTD2).<sup>1</sup> HED is the most frequent form, with an incidence of one to seven individuals per 10,000 live births.<sup>2</sup> Due to dysplasia of the ectoderm-derived structures, HED patients typically present with sparse hair, sweat gland dysplasia, dry skin, a wide and prominent forehead, saddle nose, skin pigmentation around the eyes and mouth, rough skin, increased intercanthal distance, midface depression, an upturned nose, and

mandibular protrusion.<sup>3,4</sup> As such, ED can have a detrimental impact on the function of mastication, reduce nutritional intake, and disturb the growth and development of young patients to various degrees. In addition, because of the dysmorphic features of the maxillofacial region, patients are likely to have low self-esteem, psychological pressures, and limited social interactions.

The primary dental-related features of ED patients include the congenital absence of multiple teeth, abnormal tooth shape (microdontia, conical teeth), dental hypoplasia, and abnormal tooth germination. Furthermore, the lack of tooth eruption and consequent decreased functional stimulation contribute to alveolar bone dysplasia, osteopenia, height reduction, and a knife edge alveolar ridge, which can further complicate the oral rehabilitation of these patients.<sup>5</sup>

Because ED is a genetic disorder with severe oligodontia and premature tooth loss, the oral reconstruction of ED patients is required at a young age. The reconstruction strategy is determined on the basis of many factors, including patient age, developmental stage, anatomy of the soft and hard tissues, and number of missing teeth. Therefore, the final strategies may vary significantly and include approaches such as removable partial or complete dentures, tooth-supported fixed partial dentures, overdentures, and implant-supported prostheses. However, there is increasing demand for the prostheses to restore speech, facial appearance, and comfort, and to achieve partial function of mastication. Fixed, implant-supported dentures have been used over the past 25 years in ED patients to help restore some of these complex oral functions.<sup>6</sup> However, key factors in successful implant restoration treatment depend on the quantity and quality of alveolar bone.

Due to aberrations in the craniofacial anatomy, ED patients often present with a strong desire to restore both oral function and craniofacial esthetics at an early age, to improve both the patient's social confidence and quality of life.<sup>7</sup> However, there is controversy in relation to early implant placement in children, due to their small jaw size, location/consideration of permanent tooth germs, and implant displacement with future jaw development.<sup>8,9</sup> In addition, edentulous patients often undergo extensive alveolar remodeling. As such, they present with insufficient alveolar ridge width and height. Thus, deficient alveolar ridge dimensions make it difficult to restore oral function using implant-supported prostheses. Many methodologies have been applied to augment alveolar bone in attempts to overcome the alveolar deficiencies in ED patients, including autogenous bone grafts, guided bone regeneration (GBR), maxillary sinus lifts, and distraction osteogenesis. There is limited clinical evidence from comparisons of the methodologies and outcomes of bone augmentation. Additionally, the long-term

survival and success of implants in ED patients remains unclear.

More importantly, no systematic literature review has focused on the aforementioned topics. Therefore, the aim of this systematic review was to determine the outcomes of oral reconstruction in ED patients who have received dental implant therapy through the evaluation of implant survival and success in ED patients. Many contributing parameters were also included in this analysis, such as patient age, bone grafting, implant site, and the style of the implants.

## Materials and methods

### Study selection criteria

Study inclusion criteria were as follows: (1) studies aimed at investigating ED patients with anodontia; (2) restoring the oral function of ED patients using dental implants; (3) human clinical studies including case reports, prospective and retrospective studies, and reviews; (4) at least one of the following outcomes reported: clinical, radiographic, and patient-centered; (5) reported in the English language.

Due to the lack of systematic literature reviews on this topic, broad inclusion criteria were adopted to render the findings of this review more general, without distinguishing patient characteristics, age, implant type, surgical technique, prosthetic rehabilitation, or duration of follow-up.

### Search strategy

A search was performed in the PubMed (Medline) and Web of Science databases to collect relevant information on the restoration of oral function in ED patients using dental implants. All articles published in English before October 15, 2015 were reviewed.

The article search was performed using the following key words: “(ectodermal dysplasia) AND (implant OR implants)”. Supplementary retrieval key words included the following: “ectodermal dysplasia” [MeSH term] AND “anodontia”; “ectodermal dysplasia” [MeSH term] AND “edentulous”; “ectodermal dysplasia” [MeSH term] AND “dental implants”; “ectodermal dysplasia” [MeSH term] AND “zygomatic implants”; “ectodermal dysplasia” [MeSH term] AND “implants”.

A supplementary hand search of 18 journals, mainly relevant peer-reviewed dental journals, published between 1988 and 2015, was also performed. Six journals were identified as being important to this review, namely *Oral Surgery*, *Oral*

*Medicine*, and *Oral Pathology*; *International Journal of Oral and Maxillofacial Surgery*; *Journal of Oral and Maxillofacial Surgery*; *Implant Dentistry*; *Journal of Oral Implantology*; *Pediatric Dentistry*. The reference lists of all pertinent papers and review articles were also searched for further relevant publications.

The titles and abstracts of all identified reports were analyzed by two researchers. Two independent investigators completed the preliminary screening of abstracts that were relevant to the study. The investigators then read the selected full-text articles independently, compared their selections, and resolved any conflicts in selection with a third party.

### Outcome measure

The outcome measure reported in this review was implant survival as published within each study. Implant survival refers to the presence of an implant with or without complications. Failure was defined as removal of the implant. Implant survival was calculated from the absolute number of implants placed and lost.

### Data extraction

A series of tables and figures were used to organize the clinical evidence reported in this review. The data recorded included the year of publication, the number of patients, patient age, the number of implants placed in the maxilla and mandible, type of implant, implant site, surgical technique used, prosthesis design, type of loading, follow-up period, and number of failed or lost implants.

In order to determine the methodological quality of observational and non-randomized studies, the Methodological Index for Non-Randomized Studies (MINORS) was applied to all articles to assess the quality of each study.<sup>10</sup> This index includes 12 items. The first eight items are aimed particularly at non-comparative studies and the remaining four items are used for comparative studies. Items are scored as 0 (not reported), 1 (reported but inadequate), or 2 (reported and adequate).

### Data analysis

The difference in the implant survival rate between children (age range 0–16 years) and adults with ED in selected reports was calculated using the  $\chi^2$  test. The statistical analysis of the data in this review was performed using IBM SPSS Statistics version 19.0 software (IBM Corp., Armonk, NY, USA).

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