Advice on treatment with dental implants in hypohidrotic ED

Bergendal B[1], Nielsen I[2]

[1]National Oral Disability Centre for Rare Disorders, Jönköping, Sweden

[2]Swedish ED-Society

Clinical advice for dental implant operations in children and adults with hypohidrotic ED

Individuals with ED often have dense bone quality with poor vascularisation

Be well-prepared and take the following precautions:

- Careful choice of burs, pre-tapping if necessary
- drill slowly with generous irrigation
- avoid bone substitutes if possible
- consider postoperative antibiotics

The operation should be performed by a skilled surgeon at a specialist clinic

Very young individuals with hypohidrotic ED and mandibular anodontia (total absence of primary as well as permanent teeth) have undergone dental implant treatment since the mid-1980s. Consensus conferences in Jönköping, Sweden in 1996 and 1998 advised the lowest age for treatment with dental implants in this group of patients to be around 6 years. The age of six has been recommended because the median suture, where transversal growth occurs, is normally closed at this time. This recommendation is based on analysis of growth of the mandible in children without ED. A first treatment using a removable overdenture attached to one implant in the canine region on each side of the mandible can then be performed.

In the literature there are several case reports describing successful outcomes of this kind of treatment in children. The first one reported on a Swedish boy with x-linked hypohidrotic ED[1]. A larger prospective study from the US reported on implant treatment in 51 patients with ED, both children and adults[2]. The results were presented as favourable, but on closer view the failure rates were higher than in individuals without ED.

In a questionnaire study of 109 American individuals with ED, higher rates of complications and loss of implants were reported than in studies on individuals without ED[3]. Half of the patients reported complications and 24% reported some kind of failure.

A Swedish study reported on treatment in five children with hypohidrotic ED who were treated with implants in the anodontic mandible from 5 to 12 years of age[4]. All but one boy lost one or more implants shortly after the operation. The four who lost implants were re-operated, most of them with a successful outcome. The losses of implants were attributed to the small size of the jaws and the hard bone, which made the operation technically complicated.

Later, French researchers have shown that the molecular signalling pathway in hypohidrotic ED (*EDA-EDAR-EDARADD*) leads to a transcription factor, NFκB, which affects bone and gives it a different structure and increased hardness[5]. This finding was confirmed in an Australian study where 3D-images from bone samples from young individuals with ED were compared to samples from older edentulous individuals without ED[6]. Also, individuals with incontinentia pigmenti and mutations in the *NEMO*-gene probably have harder than normal bone.

With this background, we now advise implant operations to be postponed to around 8 years of age in children missing all teeth in the lower jaw. ***The surgeon performing the operation should be prepared for the bone to be very hard.*** After growth is finished, from around 18 years of age, additional implants can be installed and a fixed dental prosthesis can be made. The treatment should then be followed by regular dental check-ups. A case-report published in 2015 provided 30 years of detailed follow-up on the successful treatment of a Swedish boy, who received his first implants at six years of age[7].

Patients with hypohidrotic ED who have some primary and/or permanent teeth can, if needed, be treated with different forms of tooth-supported removable or fixed appliances during childhood and adolescence. Treatment of these patients with dental implants can be performed after growth is almost completed in the late teens.

References

1. Bergendal T, Eckerdal O, Hallonsten AL, Koch G, Kurol J, Kvint S. Osseointegrated implants in the oral habilitation of a boy with ectodermal dysplasia: a case report. Int Dent J 1991;41:149-156.
2. Guckes AD, Scurria MS, King TS, McCarthy GR, Brahim JS. Prospective clinical trial of dental mplants in persons with ectodermal dysplasia. J Prosthet Dent 2002;88:21-25.
3. Stanford CM, Guckes A, Fete M, Srun S, Richter MK. Perceptions of outcomes of implant therapy in patients with ectodermal dysplasia syndromes. Int J Prosthodon. 2008;21:195-200.
4. Bergendal B, Ekman A, Nilsson P. Implant failure in young children with ectodermal dysplasia: A retrospective evaluation of use and outcome of dental implant treatment in children in Sweden. Int J Oral Maxillofac Implants 2008;23:520-524.
5. Lesot H, Clauss F, Manière MC, Schmittbuhl M. Consequences of X-linked hypohidrotic ectodermal dysplasia for the human jaw bone. Front Oral Biol 2009;13:93-99.
6. Silthampitag P, Klineberg I, Jones AS, Austin B, Zee KY, Wallace C, Scholz S, Naim A, Zoud K. Ultramicroscopy of bone at oral implant sites: a comparison of ED and control patients. Part 1-defining the protocol. Int J Prosthodont. 2011;24:147-154.
7. Bergendal B, Bjerklin K, Bergendal T, Koch G. Dental Implant Therapy for a Child with X-linked Hypohidrotic Ectodermal Dysplasia - Three Decades of Managed Care. Int J Prosthodont 2015;28:348-356.

2016-02-22

Birgitta Bergendal, PhD, specialist in prosthetic dentistry
National Oral Disability Centre for Rare Disorders
The Institute for Postgraduate Dental Education, Jönköping
http://plus.rjl.se/oi

Ia Nielsen
Karlskoga
Swedish ED-society
http://www.svenskaed.se