Interpretive and Report Bias in Publications on Implants in Patients with Ectodermal Dysplasia

The clinical scientific community's acceptance of the efficacy and effectiveness of osseointegrated dental implants has led to the challenging exploration of possible uses of the method in other groups of patients. One of the strongest indications for early treatment was anodontia of the mandible in young boys with hypohidrotic ectodermal dysplasia (HED). The first two cases were treated in the mid-1980s, one in Sweden and one in California, and both reported favorable results.[1,2] Two implants were placed in the canine region of the mandible to support an overdenture. The boy treated at our center has been followed for over 25 years, and we have reported it in different fora as a success story.

In 1996, a consensus conference on oral implants in young patients was held in Jönköping, Sweden,[3] followed by a conference on ED in 1998.[4] In the book from the latter conference, five clinical cases were presented, three of which were boys with HED and an anodontic mandible; one case was the boy first reported from Sweden, and the other two had lost implants shortly after they were placed. However, at that time, these implant failures were attributed to the overall patient management and were not interpreted to be related to the diagnosis.

A compilation on the use of dental implants in individuals with rare disorders found 57 publications reporting on 151 patients.[5] More than half of the publications reported on patients with ED, and more than 70% of patients had ED. Of 31 publications on ED, 19 were individual cases reporting successful treatments; only one was a prospective clinical trial reporting on 51 individuals aged 8 to 68 years who had 251 implants placed.[6] Under the heading "Clinical Implications," the following was stated: "This short-time study demonstrated that implant osseointegration can be successful in subjects with varying ages with severe hypodontia."[6]

In 2005, there were reports to our center from the Swedish ED Society that some young children had lost implants shortly after placement. To further explore this information, a survey was sent to all Swedish specialist clinics in oral and maxillofacial surgery and prosthetic dentistry asking them to provide results of dental implant treatment in children up to age 16. In all, 26 patients were reported who were treated from 1996 to 2005; 5 had HED. All patients with HED except for the first boy previously mentioned had lost implants before loading, and the failure rate was 64% (9 of 14 implants).[7] In a discussion with the oral surgeons who had performed the operations, all described difficulties related to the small bone volume and extremely hard bone. Illustrated by superimposing an implant on a computed tomography scan from a small child with anodontia, a risk that the implant was placed in cortical bone along the buccal and lingual aspects of the mandible was visualized. However, all 4 cases with failures had successful implant placements at a later point, the first 2 patients when they were in their teens and the other 2 immediately after primary healing.

The conclusion in our study was that preoperative conditions, ie, the small size of the jaws and the hard bone, and not the syndrome per se made implant treatment difficult and increased the risks for failure in small children with HED.

In retrospect, a similar failure rate was seen in the first three cases reported at the conference in 1998, where five of eight implants (62.5%) were lost. A compilation of four studies on implant treatment in groups of individuals with ED showed that early failures were reported in the mandible by Kearns et al,[8] Guckes et al,[6] and Sweeney et al,[9] with failure rates ranging from 2.4% to 9.1%.

More recently, Lesot et al demonstrated increased jawbone density in patients with X-linked HED, and concluded that the skeletal phenotype is associated with the mutation while confirming the involvement of the EDA-NF-$_k$B pathway in bone metabolism.[10] In a comparison of jawbone in young individuals with ED and unaffected adult controls, bone samples harvested using a trephine bur at implant sites were examined using a SkyScan x-ray microtomography system.[11] The analyses showed that bone from the ED group had a denser, more compact, and well-connected structure. This is an intriguing result showing a difference in bone density and structure in individuals with ED, even if a recent review found no studies that directly related bone density to implant survival.[12]

The high failure rates in young Swedish children with HED have been referred to as follows: "These

disappointing results correspond with unsubstantiated clinical observations of compromised and unpredictable survival of dental implants in other patient groups with severe hypodontia, which is often attributed to 'poor bone quality.'"[13] On more than one occasion when I have reported on implant failures in children with HED, people from different parts of the world have come up afterward and told me of similar experiences of failed implants in patients with HED. There is reason to believe that predominantly successful cases have been published, which might have postponed our understanding of specific risks related to implant treatment in patients with HED.

Thus, though delayed by interpretive as well as report bias, a picture is emerging of a higher risk of implant failures in individuals with HED. Clinical implications from current knowledge could be a recommendation not to place implants in children with HED as young as 6 years of age and an anticipation of the risks in handling very dense bone.

The reporting of complications and failures is of utmost importance in the pursuit of specific risks related to certain diagnoses. This is even more crucial in rare disorders where certain types of treatment are performed less frequently. The reporting of failed implants in individuals with rare disorders is therefore strongly advocated. Quality registers and multicenter cooperation would open up possibilities to prospectively monitor outcomes of treatment and indicate risks and adverse effects. Moreover, these concerns underscore an articulated emphasis on the likelihood that both early and late failure to osseointegrate may result from variations in an individual's systemic or even specific host bone sites' healing potential.[14]

Birgitta Bergendal, DDS, PhD
National Oral Disability Centre
The Institute for Postgraduate Dental Education
PO Box 1030
SE 551 11
Jönköping, Sweden
birgitta.bergendal@lj.se

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. Smith RA, Vargervik K, Kearns G, Bosch C, Koumjian J. Placement of an endosseous implant in a growing child with ectodermal dysplasia. Oral Surg Oral Med Oral Pathol 1993;75: 669–673.
3. Koch G, Bergendal T, Kvint S, Johansson U (eds). Consensus Conference on Oral Implants in Young Patients. The Institute for Postgraduate Dental Education, Jönköping, Sweden. Stockholm: Förlagshuset Gothia, 1996.
4. Bergendal B, Koch G, Kurol J, Wänndahl G (eds). Consensus Conference on Ectodermal Dysplasia with Special Reference to Dental Treatment. The Institute for Postgraduate Dental Education, Jönköping, Sweden. Stockholm: Förlagshuset Gothia, 1998.
5. Bergendal B, Anderson JD, Müller F. The challenging patient with facial deformities, rare disorders, or old age. In: Jokstad A (ed). Osseointegration and Dental Implants. Ames, Iowa: Wiley-Blackwell, 2009:43–62.
6. Guckes AD, Scurria MS, King TS, McCarthy GR, Brahim JS. Prospective clinical trial of dental implants in persons with ectodermal dysplasia. J Prosthet Dent 2002;88:21–25.
7. 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.
8. Kearns G, Sharma A, Perrott D, Schmidt B, Kaban L, Vargervik K. Placement of endosseous implants in children and adolescents with hereditary ectodermal dysplasia. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1999;88:5–10.
9. Sweeney IP, Ferguson JW, Heggie AA, Lucas JO. Treatment outcomes for adolescent ectodermal dysplasia patients treated with dental implants. Int J Paediatr Dent 2005;15:241–248.
10. Lesot H, Clauss F, Maniére MC, Schmittbuhl M. Consequences of X-linked hypohidrotic ectodermal dysplasia for the human jaw bone. Front Oral Biol 2009;13:93–99.
11. Silthampitag P, Klineberg I, Jones AS, et al. 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.
12. Martin W, Lewis E, Nicol A. Local risk factors for implant therapy. Int J Oral Maxillofac Implants 2009;24(suppl):28–38.
13. Créton M, Cune M, Verhoeven W, Muradin M, Wismeijer D, Meijer G. Implant treatment in patients with severe hypodontia: A retrospective evaluation. J Oral Maxillofac Surg 2010;68: 530–538.
14. Chvartszaid D, Koka S. On manufactured diseases, healthy mouths, and infected minds. Int J Prosthodont 2011;24:102–103.

Birgitta Bergendal had her specialty training at the Karolinska Institute in Stockholm, Sweden, and holds a doctoral degree from Umeå University, Umeå, Sweden. In 1982, she joined the Department of Prosthetic Dentistry at the Institute for Postgraduate Dental Education in Jönköping. She then became head of the National Oral Disability Centre for rare disorders in 1999. The center was established to increase knowledge about the diagnosis and treatment of oral and dental signs and symptoms of rare disorders.