Implant Failure in Young Children with Ectodermal Dysplasia: A Retrospective Evaluation of Use and Outcome of Dental Implant Treatment in Children in Sweden

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Purpose: *This study surveyed dental implant treatment in children up to age 16 years in Sweden between 1985 and 2005, with special reference to young children with ectodermal dysplasia (ED) and anodontia in the lower jaw.* ***Materials and Methods:*** *A questionnaire was sent to Swedish specialist clinics in oral and maxillofacial surgery and prosthetic dentistry. Also, the teams who had treated children with ED were asked to submit their records for these children for a discussion on reasons for implant failure.* ***Results:*** *Six out of 30 specialist centers (20%) in Sweden had treated 26 children with dental implants between 1985 and 2005. Twenty-one patients had received 33 implants to replace teeth missing from nonsyndromic agenesis or trauma at ages 14 or 15 years; 2 (6.1%) of these implants were lost. Five children with ED received 14 implants at 5 to 12 years of age; 9 (64.3%) of these implants were lost before loading.* ***Conclusions:*** *Dental implant placement has been a rarely used treatment modality in Swedish children less than 16 years old in the last 20 years. The failure rate in children treated because of tooth agenesis was only slightly higher than that reported for adult individuals, whereas in young children with ED and anodontia in the mandible, implants seemed to present special challenges, and the failure rate was very high. The small jaw size and peroperative conditions, rather than ED per se, were thought to be the main risk factors. Centralizing implant operations in young children with ED and monitoring outcomes in implant registers are strongly advocated.*
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Key words: *children, dental implants, ectodermal dysplasia, failure, rare disorders*

Dental implant treatment has rarely been used in growing individuals, apart from in children with ectodermal dysplasia (ED) syndromes and anodontia in the mandible, where early treatment has been recommended.[1] Several case reports of successful treatment with implant-supported overdentures in the mandibles of 3- to 6-year-old boys with x-linked hypohidrotic ED have been published in the last 15 years.[2–4] The youngest child treated was 1.5 years old.[5]

A Scandinavian consensus conference on dental treatment of children with ED was held in Jönköping, Sweden, in 1998. A care program was drawn up, where oral habilitation, including treatment with dental implants in anodontic mandibles, was recommended before school age (6 to 7 years). The outcomes of dental implant treatment in 3 cases of young children with hypohidrotic ED were presented in detail in the conference publication.[6] All 3 patients had mandibular anodontia and received 2 to 4 implants under general anesthesia. The implants placed in 1 of the patients were successful, while the other two patients experienced early loss of implants. In discussions during the conference, lack of patient monitoring following surgery and orofacial motor dysfunction were factors thought to have negatively influenced osseointegration.

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Table 1 No. of Children Treated with Dental Implants in Sweden Between 1985 and 2005

Diagnosis	Gender (girl/boy)	Birth year	Age at operation (y)	Implants placed (n)	Implant location (maxilla/mandible)	Implants lost N (%)
Agenesis	13/1	1973–1983	14–15	25	11/14	2 (8.0%)
Trauma	4/3	1975–1982	12–15	8	7/1	0
ED	1/4	1979–1999	5–12	14	0/14	9 (64.3%)
Total	26			47	18/29	11 (23.4%)

ED = Ectodermal dysplasia.

Table 2 Children with Hypohidrotic Ectodermal Dysplasia and Anodontia in the Mandible Who Received Dental Implants Between 1985 and 2005

Patient	Year of operation	Age at operation	Gender	Implants placed (n)	Implants lost (n)	Type of implant
1	1985	6	Boy	2	0	Brånemark System Standard implant Ø 3.75 10 and 13 mm long
2	1988	6	Boy	4	3	Brånemark System Standard implant Ø 3.75 Three 13-mm-long implants, one 10-mm-long implant
3	1991	8	Boy	2	2	Brånemark System Standard implant Ø 3.75 × 10 mm
4	2004	12	Girl	4	3	Brånemark System Mk III TiUnite NP Ø 3.3 × 13 mm
5	2005	5	Boy	2	1	Brånemark System Mk III TiUnite RP Ø 3.75 × 10 mm
Total				14	9	

NP = narrow platform; RP = regular platform.

In 2005 the National Oral Disability Centre in Jönköping received reports from the Swedish patient organization for ectodermal dysplasias that some recently treated young patients had experienced complications and implant loss. It was then decided, in cooperation with the Swedish National Board of Health and Welfare and the Swedish ED Society, to retrospectively evaluate the use and outcome of dental implant treatment in young individuals in Sweden. Sweden has 9 million inhabitants, and dental treatment, including dental implants, is free of charge up to the age of 19 years. Treatment with dental implants is a frequently used treatment modality in adults, and implant operations in the edentulous mandible is a routine operation for specialists in oral and maxillofacial surgery.

MATERIALS AND METHODS

In September 2005, a questionnaire was sent to Swedish specialist clinics in oral and maxillofacial surgery and prosthetic dentistry requesting information on all children who had been treated with dental implants. The inclusion criterion was age below 16 years at the time of the implant operation. A simple form was used for each patient; the form comprised questions on age and gender, reason for treatment, syndrome diagnosis, number of implants placed in the maxilla and mandible, and number of lost implants. After analysis of the results, 5 teams who had treated children with hypohidrotic ED were asked to send clinical records (operation record, radiographs, and clinical photographs) about the treatment. These were distributed to 3 oral and maxillofacial surgeons, who then participated in a telephone conference to discuss reasons for implant failure.

RESULTS

Forty-two clinics representing 30 specialist centers throughout Sweden responded. Six centers (20%) had treated 26 children below age 16 (18 girls and 8 boys) with dental implants between 1985 and 2005. Reasons for treatment were nonsyndromic tooth agenesis (n = 14), trauma (n = 7), and ED and anodontia in the mandible (n = 5; Table 1). In the agenesis group, implants were placed in the maxilla and mandible; in the trauma group, mostly in the maxilla; and in the ED group, only in the anterior region of the anodontic mandible.

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Fig 1 Situation at implant operation. Note exposed threads on left implant (patient 1).

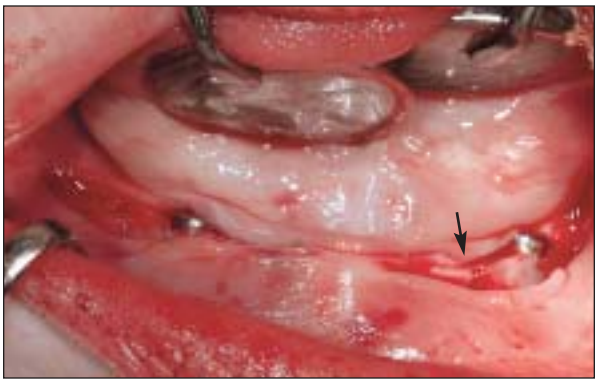

Fig 2 Situation at second implant operation after loss of an implant that had been placed anteriorly on the left side (patient 5). Arrow indicates first implant site with buccal fracture.

In the agenesis and trauma groups, age at implant operation was 14 or 15 years, except for one 12-year-old boy who had received an implant to replace a maxillary central incisor lost from trauma. This was published in 1994 as the first case report of a follow-up of a single-tooth implant in a child.[7] In the agenesis and trauma groups, 33 implants were placed and 2 (6.1%) were lost. The 5 children with ED were treated at 4 centers by 5 teams of specialists. The first patient in the ED group underwent surgery in 1985, had no complications, and was presented as a case report in 1991.[2] The 5 patients with ED received 14 implants and lost 9 (64.3%) before loading. Table 2 lists each patient and type of implant in the ED group.

The records of the 5 patients with ED clearly showed that the dimensions of the mandible were very limited at the time of operation. This was commented upon in the case presentation of the first patient,[2] where 4 threads of 1 of the implants were exposed on the buccal side at the time of implant placement (Fig 1).

After their discussion, the surgeons reported that they considered the cortical bone in the last 2 patients, treated in 2004 and 2005, to be very dense. Bone quality was estimated to be 1 and 1.5, respectively, according to the criteria established by Lekholm and Zarb.[8] In 1 patient, the implants could not be placed with an implant inserter but were instead screwed in by hand with great force. Shortly after primary healing, fistulas developed, and the implants were lost within 6 weeks. In the other patient, a 5-year-old boy, the buccal bone fractured at one implant site during the implant operation, and the implant was lost spontaneously after 2 months (Fig 2).

Brånemark System implants (Nobel Biocare, Göteborg, Sweden), 10 or 13 mm in length and with varying diameters and surface structures, were placed in all 5 patients with ED. The patient who suffered no implant loss had undergone a prolonged healing time of 6 months before the abutment operation. In the other 4 patients, the implants were lost before abutment connection. In these patients, new implants were successfully placed later. In patients 2 and 3, more implants were placed at the ages of 15 and 19 years, respectively, and patients 4 and 5 underwent replacement operations within the same year after initial healing.

The major risk factor agreed upon in the surgeons' discussion was the low quantity of bone, that is, the small dimensions of the mandible in the small children. They also discussed the patients' bone quality, estimations of which emanated from feel during surgery. The cortical bone was considered to be very dense, and the cancellous bone was described as very loose. To better understand the dimensional challenges, a data simulation was made from a computed tomography (CT) scan of the canine region of the mandible of an edentulous adult person. The true size of the height of the bone on the digital CT scan was calculated. From an assumption that the difference in body height between an adult and a 6- to 8-year-old child on average exceeds at least one-third of an adult's height, the size of the CT scan was scaled down by 30%. Digital images of 10-mm regular-platform implants 3.75 mm in diameter (Fig 3) and 10-mm narrow-platform implants 3.3 mm in diameter (Fig 4; Brånemark System) were superimposed on this CT scan. This illustrated the small buccolingual dimensions and the small amount of cancellous bone around the implant. In some cases, the entire length of the implant may have been in contact with cortical bone.

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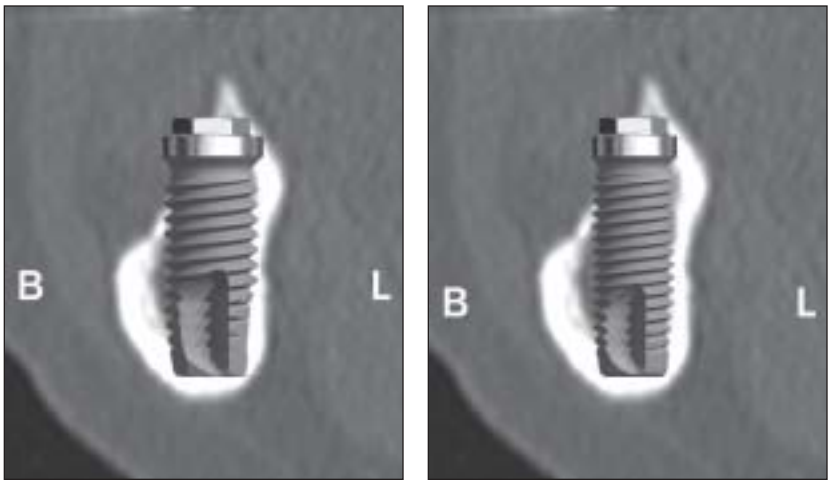

Fig 3 *(Left)* Tomogram (size reduction 30%) of the canine region of an edentulous adult with a superimposed Brånemark System TiUnite implant, regular platform, 3.75 mm wide and 10 mm long.

Fig 4 *(Right)* Tomogram (size reduction 30%) of the canine region of an adult edentulous individual with a superimposed Brånemark System TiUnite implant, narrow platform, 3.3 mm wide and 10 mm long.

DISCUSSION

Ödman[9] established that an implant does not follow the appositional growth of the alveolar process but behaves like an ankylosed tooth. Johansson et al[7] published a case report in 1994 of placement of a dental implant in a 12-year-old boy at the site of a maxillary central incisor lost due to trauma. The implant was followed for 4.5 years, and infraocclusion of the implant-supported crown was clearly demonstrated. This was the background for the recommendation made in the consensus statements at the 1996 conference in Jönköping, Sweden, to postpone dental implant treatment in children until well after the pubertal growth spurt.[1]

That an implant behaved like an ankylosed tooth was later clearly illustrated by Rossi and Andreasen in 2003 in the case report[10] of a boy who had received an implant at the site of a maxillary central incisor at age 10 and was followed for 10 years. The implant-supported crown was exchanged several times, and the distance between the head of the implant and the cementoenamel junction of the neighboring tooth was 10 mm. However, in the case of anodontia of the mandible, the consensus statements from the 1996 conference on implants in children[1] and the 1998 conference on dental treatment in ED[11] recommended early treatment with dental implants. Regarding oral rehabilitation of patients with ED,[6] 2 cases were presented at the 1996 conference where implants were lost in the anodontic mandibles of young boys with hypohidrotic ED. Guckes et al[12] have published the only prospective study on dental implants in persons with ED. Of the 46 implants placed in children less than 11 years old (the youngest age group), 6 implants (13.0%) were lost in 3 individuals. To date, no further indications of complications or implant loss in young children with ED have been published. From the follow-up of the first patient, treated more 20 years ago, it can be concluded that treatment of a growing child with implant-supported overdentures requires a team of dedicated specialists who are responsible for regular follow-ups and for the relining, rebasing, and remaking of the dentures.

This retrospective study found that 47 implants had been placed in 26 children during a 20-year period in Sweden and that only 1 in 5 specialist centers had treated children under age 16. Even though the material comprises only a few young patients, the failure rate of implant treatment in children in cases of tooth agenesis appears to be slightly higher than of that of treatment with single-tooth implants in adult individuals.[13] No implants were lost in the group treated due to frontal trauma, predominantly in the maxilla. All children but 1 in these 2 groups were 14 or 15 years old at the time of operation. The children with ED were much younger; 4 were 5 to 8 years old and 1 was 12. The small size of the jaws is presumed to be the predominant reason for implant failure; the surgeons may have been unprepared for the technical difficulties that this implied.

At sites where there has never been a tooth, neither primary nor permanent, the prerequisites for bone healing and osseointegration have been postulated to be different. However, no studies were found to support the hypothesis that lower cell activity negatively affects bone healing and osseointegration. On the contrary, the favorable outcomes of treatment of young children with cochlear implants in the mastoid process, where cancellous bone is usually very loose and the chances of achieving primary stability are often compromised, demonstrate that these risk factors can be overcome by good surgical technique.[14] Also, short 6- and 7-mm Brånemark implants that were placed in adults with severely atrophic edentulous mandibles and were almost entirely in contact with cortical bone were reported to have a cumulative implant survival rate of 95.5% after 5 years.[15]

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Experience in implant treatment of young children with ED is still limited. Since all 4 patients had successful reoperations, the high number of lost implants in this Swedish study indicated that peroperative conditions rather than ED per se were important reasons for the failures. The successful case reports might have given the impression that placing implants in anodontic mandibles of young children with ED is uncomplicated. Recent results seem to emphasize that implant surgery in this group of patients presents special challenges, mainly because of the small dimensions of the jaws and the dense cortical bone.

Strategies for minimizing implant loss in this group of patients include a CT examination combined with thorough treatment planning, the use of small-diameter implants, and postponement of treatment until jaw size is sufficient to support an implant. Well-known surgical principles of atraumatic surgery, the use of a screw tap before implant insertion, and excessive cooling during the implant operation are techniques that must be used. From an ethical standpoint, treatment of small children under general anesthesia requires special attention to possible risks and side effects. Implant treatment in small children therefore ideally ought to be centralized to create specialist teams whose collected experience has a chance to grow. Multicenter studies of cleft lip and palate treatment clearly demonstrated that centralization and high-volume operators were associated with better outcomes.[16] Centralizing oral rehabilitation in the treatment of individuals with ED and other rare disorders would also create the best possible evidence base for choice of treatment. Setting up quality registers to monitor outcomes of dental implant treatment in children could promote early detection of risky situations.

CONCLUSIONS

Dental implant treatment has been a rarely used treatment modality in young children in Sweden in the last 20 years. Nine of 14 implants placed in children with ectodermal dysplasia and anodontia in the mandible were lost before loading. The small dimensions of the jaws and peroperative conditions rather than the ectodermal dysplasia per se were interpreted to be the main reasons for implant failure. Choosing implants that are well suited for the small size of the jaws and postponing treatment for a few years are strategies that can be recommended to minimize the risk of implant loss. Centralizing the oral rehabilitation of individuals with ED and other rare disorders and monitoring outcomes are generally advocated.

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