Growth Analysis of a Patient with Ectodermal Dysplasia Treated with Endosseous Implants: A Case Report

Karin B. Becktor, DDS[1]/Jonas P. Becktor, DDS[2]/Eugene E. Keller, DDS, MSD[3]

Congenital absence of multiple teeth and poorly developed alveolar ridges are associated with ectodermal dysplasia. Affected patients often require dental prosthetic treatment during their developmental years. Maxillofacial growth and development in a preadolescent female patient with ectodermal dysplasia following oral rehabilitation with maxillary and mandibular endosseous dental implants is reported. Four maxillary and 4 mandibular implants were successfully integrated and restored at 8 years of age. Growth analysis 12 years later revealed that the implants followed maxillary and mandibular growth displacement. Minor impaction of the maxillary implants was observed, and mandibular implants were affected by the mandibular growth rotation, which led to a change in implant inclination. The treatment outcome is compared to similar previously reported studies and cases. (INT J ORAL MAXILLOFAC IMPLANTS 2001;16:864–874)

Key words: *craniofacial growth, ectodermal dysplasia, endosseous dental implants*

Ectodermal dysplasia represents a group of inherited conditions that adversely affect ectodermal structures such as hair, skin, nails, and teeth. The frequency of hypohidrotic ectodermal dysplasia (HED) is about 1/100,000.[1] Congenital missing primary and permanent teeth in the maxilla and mandible and associated hypoplasia of the alveolar bone are the most frequently described oral abnormalities.[2] The number of missing teeth varies, with a higher incidence in the mandible. When present, teeth are frequently deformed; crowns are conical and taurodontism (prism-shaped molars with large pulp chambers) is common. In the maxilla, the most frequently existing primary teeth are second molars, canines, and central incisors; in the permanent dentition, first molars, canines, and central incisors are the most commonly present. In the mandible, primary canines followed by permanent canines and first molars are the most frequently present.[2] The etiology for this type of dental agenesis is dysplasia of the oral epithelium.[3]

Clinical management of oligodontia presents many problems for the restorative dentist. Removable partial or complete dentures have been proposed as the treatment of choice in the affected growing individual. Currently, endosseous dental implants are considered the ideal reconstruction modality following completion of alveolar growth and development. A retentive and stable removable partial or complete denture in preadolescent ectodermal dysplasia patients is difficult to achieve because of the absence of alveolar bone and continuous changing of maxillary and mandibular basal bone relationships. To avoid functional and/or psychosocial compromise, early treatment with dental endosseous implants is a worthy goal in these patients.

[1]Visiting Clinician, Division of Orthodontics, Dental Specialties, Mayo Clinic, Rochester, Minnesota; PhD Candidate, Department of Orthodontics, School of Dentistry, University of Copenhagen, Denmark.

[2]Visiting Clinician, Department of General Surgery, Division of Oral and Maxillofacial Surgery, Mayo Clinic, Rochester, Minnesota; Senior Registrar, Department of Oral and Maxillofacial Surgery, Länssjukhuset, Halmstad, Sweden.

[3]Consultant, Department of General Surgery, Division of Oral and Maxillofacial Surgery, Mayo Clinic, and Professor, Mayo Medical School, Rochester, Minnesota.

Reprint requests: Dr K. B. Becktor, Department of Orthodontics, School of Dentistry, Faculty of Health Sciences, University of Copenhagen, 20 Nørre Allé, DK-2200 Copenhagen N., Denmark. Fax: +45-35-326505. E-mail: kbb@odont.ku.dk

The treatment outcome in a female patient with hypohidrotic ectodermal dysplasia and oligodontia was analyzed 12 years after the placement of endosseous dental implants in the posterior maxilla and anterior mandible at age 8.

PATIENT HISTORY

The patient was a female, the third child of parents of unknown genetic background. She was born as a "normal" infant at term weighing 7 lbs, 4 oz after a normal pregnancy. One male and one female sibling were described as normal. The mother had experienced spontaneous miscarriage on 2 occasions. In the neonatal period, the mother noticed that the child was sweating very little and was heat-intolerant. Academic progress was judged to be normal, and the patient's health in general was described as excellent.

Physical Examination

A comprehensive examination of the patient at age 8 years, 7 months was undertaken at the Mayo Clinic (Rochester, MN) by a multidisciplinary team that included representatives from oral and maxillofacial surgery, prosthodontics, dermatology, and genetics.

Physical Appearance. The patient had diffusely sparse head hair, eyebrows, and eyelashes, and no patches of alopecia (loss of hair). She had severe hypohidrosis (diminished sweating), dry skin, and dyschromasia (abnormal pigmentation) in the orbital region. The nails were normal and the girl was otherwise healthy.

Genetics. The father had several congenitally missing teeth and mild hypohidrosis. The milder expression in the father, compared to the severe expression in the daughter, argues strongly against an x-linked inheritance. Moreover, the patient's sister had no sign of ectodermal dysplasia. If the pattern of inheritance had been x-linked dominant, at least some expression in the sister would have been expected. Therefore, the diagnosis of hypohidrotic ectodermal dysplasia with autosomal dominant inheritance was given.

Oral Examination. The patient had severe oligodontia. The only teeth present were the maxillary primary canines and permanent mandibular first molars (Fig 1). The canine crowns had a conical shape. The alveolar process in the edentulous regions was hypoplastic.

Fig 1 Dentition at age 8 years. The only developed teeth are the maxillary primary canines and mandibular first molars.

Radiologic Investigation

Panoramic Radiographs. Figures 2a to 2d show the dentition and bone appearance from age 8 years, 7 months to age 20 years. The first molars were taurodontic and the alveolar process was hypoplastic in all edentulous areas. Vertical alveolar growth occurred with the development and eruption of the maxillary canines and mandibular first molars. The relative vertical position of the maxillary posterior implants changed relative to the nasal floor (Figs 2b and 2c), as the result of surface remodeling of the nasal floor.

Lateral Cephalometric Radiographs. Table 1 shows the craniofacial morphologic analysis, and the reference points and lines used for analysis are shown in Fig 3. Before treatment, reduced vertical face height, a retrognathic maxilla, and lack of alveolar development in the edentulous areas characterized the morphology. After implant placement, there was mandibular backward (clockwise) rotation type I, a reduced mandibular plane angle, and an overall increase in vertical face height (Table 1).

Treatment Objectives

Because of the functional and psychosocial needs of the patient, dental endosseous implants were considered a reasonable treatment option in spite of her young age.

Surgical Management

At age 8 years, 8 months, four 10-mm self-tapping threaded cylindric endosseous implants were placed in the maxillary left and right canine and first premolar regions. The alveolar process was short and narrow. Basal bone was present but marginal in width and height. The 4 Brånemark System implants (Nobel Biocare, Göteborg, Sweden) extended into the piriform rim cortex anteriorly and into the anterior antral cortex posteriorly.

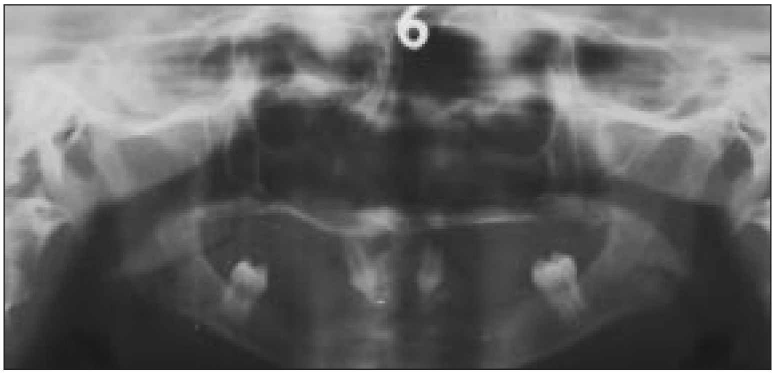


Fig 2a Orthopantomogram at age 8 years.

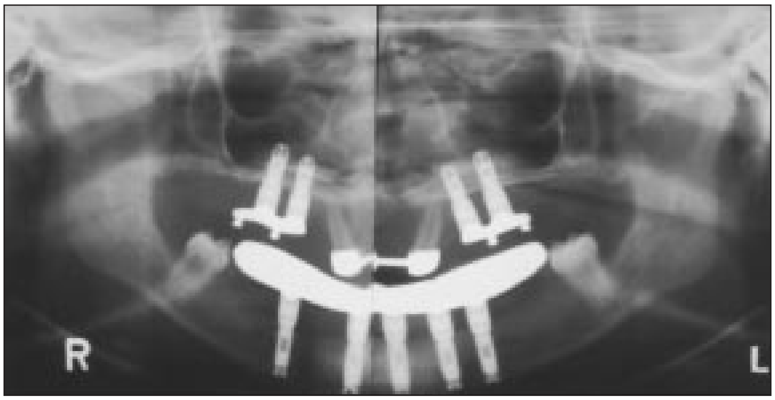


Fig 2b Orthopantomogram at age 9 years, 11 months after placement of endosseous implants.

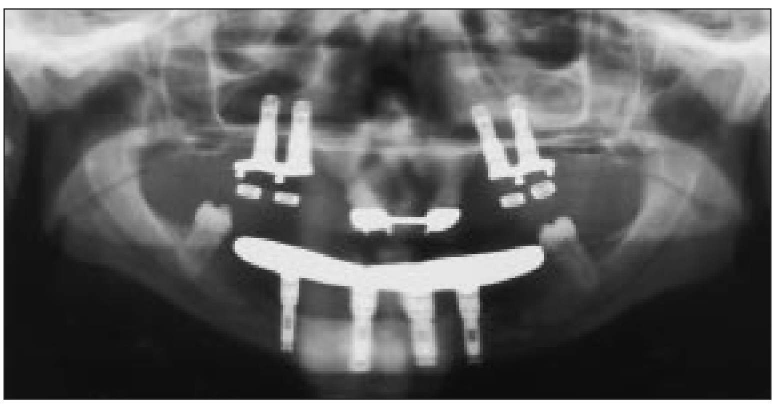


Fig 2c Orthopantomogram at age 19 years. Note the eruption of the mandibular first molars.

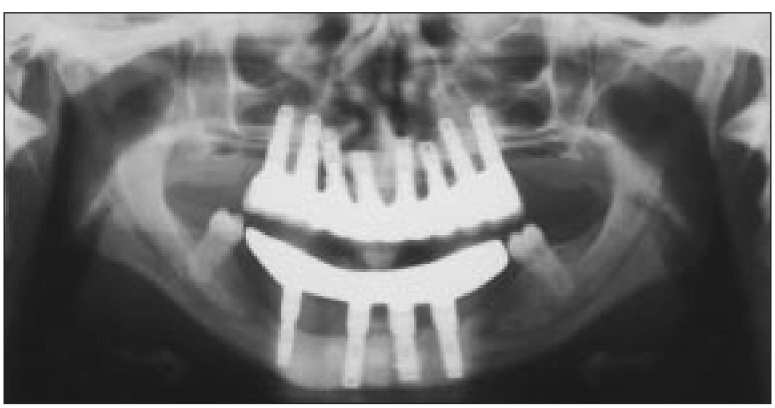


Fig 2d Orthopantomogram at age 20 years. The canines were extracted because of caries and periodontal bone loss. Four endosseous implants were placed in the anterior maxilla. The 8 implants that had been placed 12 years earlier were still clinically and radiographically osseointegrated.

Table 1 Craniofacial Cephalometric Analysis

Age	SNA angle (deg)	SU*	S-N-Pog (deg)	SU*	A-N-Pog (deg)	SU*	AN/PP (deg)	SU*	SN/MP (deg)	SU*	N-S-Ba (deg)	SU*	Inclination of lower implant to mandibular plane
8 y, 10 mo	72.0	–2.9	78.0	–0.6	–6.0	–3.2	4.0	–1.3	27.0	–1.0	131	0	—
9 y, 7 mo	71.5	–3.3	75.0	–1.4	–3.5	–2.2	3.0	–1.7	39.5	1.1	131	0	70.0
20 y	72.0	–2.9	81.0	–0.3	–9.0	–4.4	3.5	–1.5	33.5	0.1	126	–1.1	68.5

*$\frac{x-\bar{x}}{SD}$ = SU (standard units), where x is the observation and $\bar{x}$ is the mean.

S = sella; N = nasion; A = point A; Pog = pogonion; SN = sella-nasion line; PP = palatal plane; MP = mandibular plane; Ba = basion.

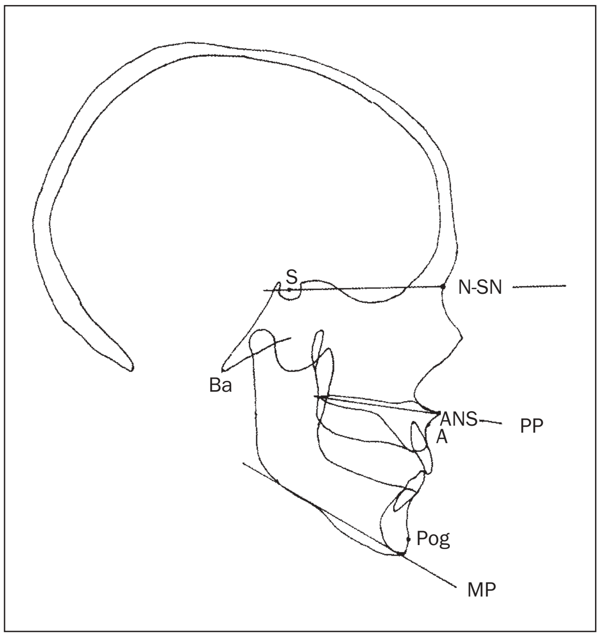


Fig 3 *(Left)* Skeletal reference points and lines. For definitions see Solow.[25] A = point A; Ba = basion; MP = mandibular plane; N = nasion; Pog = pogonion; PP = palatal plane; S = sella; SN = sella-nasion line; ANS = anterior nasal spine.

Fig 4 The definitive fixed implant-supported maxillary and mandibular prostheses at age 20 years.

In the mandible, the alveolar process was also short and narrow. The basal bone was of typical size for an edentulous mandible in a nearly 9-year-old patient. Bone density was normal in both jaws. Four 13-mm standard and one 15-mm standard Brånemark system implants were placed between the mental foramina. Abutment connection surgery was performed after 10 months of healing. One nonintegrated mandibular implant was removed.

The patient returned 10 years later at age 19 years for further prosthetic treatment, since the fixed/removable maxillary prosthesis was unstable and did not relate well to the fixed mandibular prosthesis. The maxillary canines were extracted because of horizontal bone loss and caries. Two 10-mm and two 13-mm endosseous implants were then placed in the anterior maxilla and penetrated the anterior nasal floor cortex. Thirteen months later, abutment connection surgery was accomplished. Abutments on the 4 previously placed maxillary implants were lengthened from 3 to 7 mm.

Prosthetic Treatment

At age 9, the patient was provided with a maxillary fixed/removable prosthesis supported by the bilateral posterior implants and the anterior canine teeth. The mandibular prosthesis was of standard fixed-type fabrication. At age 20 years, fixed implant-supported prostheses were placed in both the maxilla and the mandible (Fig 4).

Growth Analysis

Growth analysis from age 9 to 20 years showed that the implants in both the maxilla and mandible

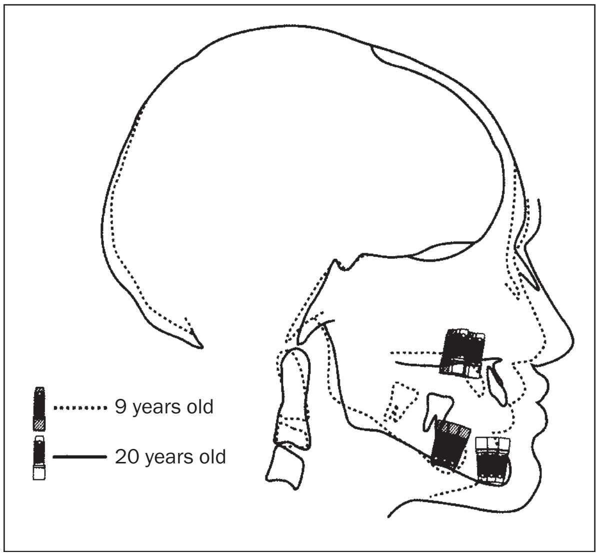


Fig 5a Superimposition on stable structures in the anterior cranial base from age 9 years to age 20 years.

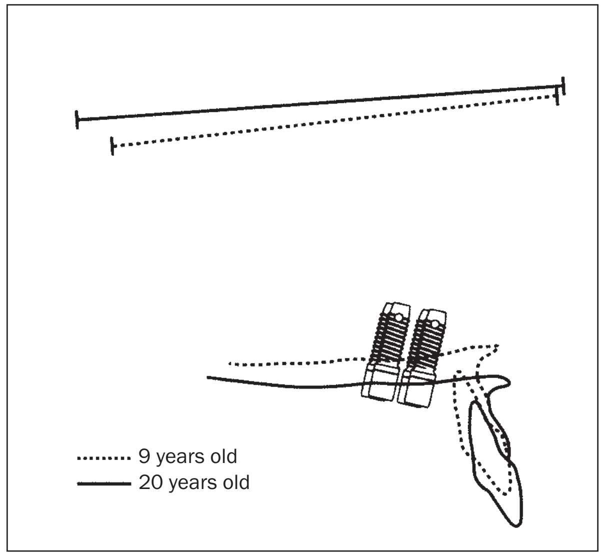


Fig 5b Maxillary superimposition on stable structures and endosseous implants during the same period, according to Björk and Skieller.[19] Note relocation of the nasal floor.

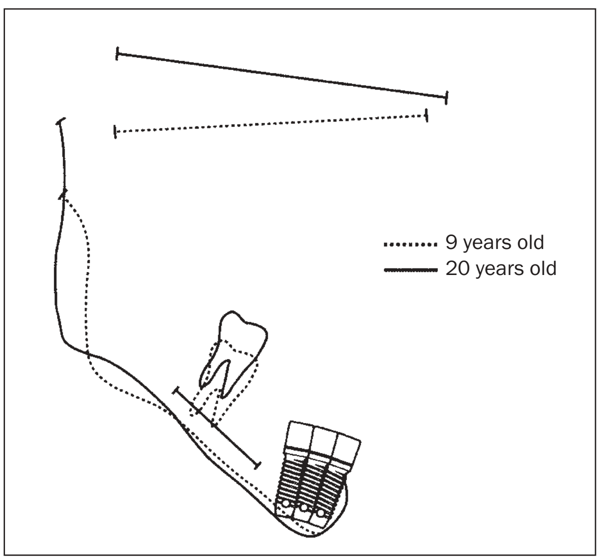
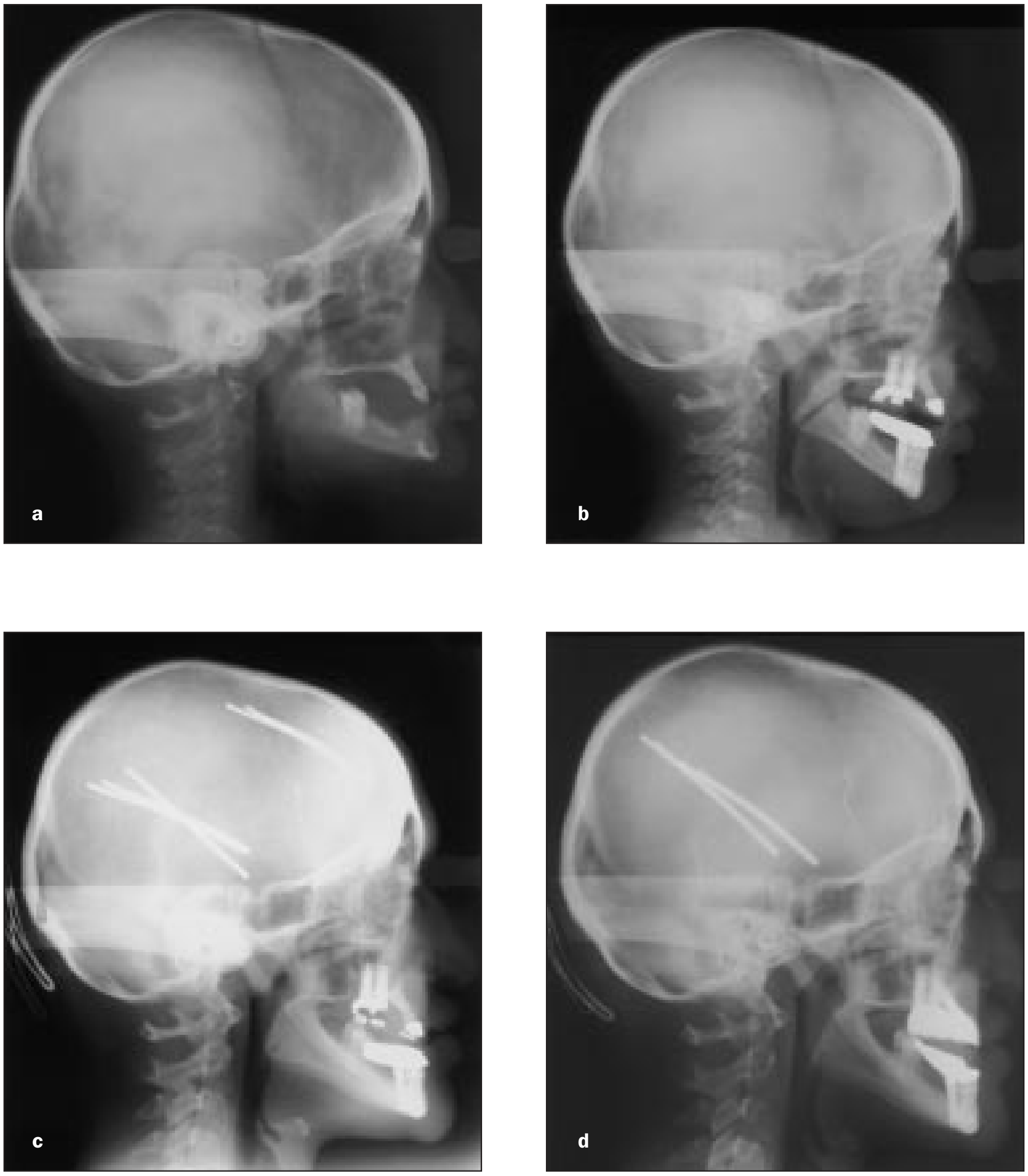


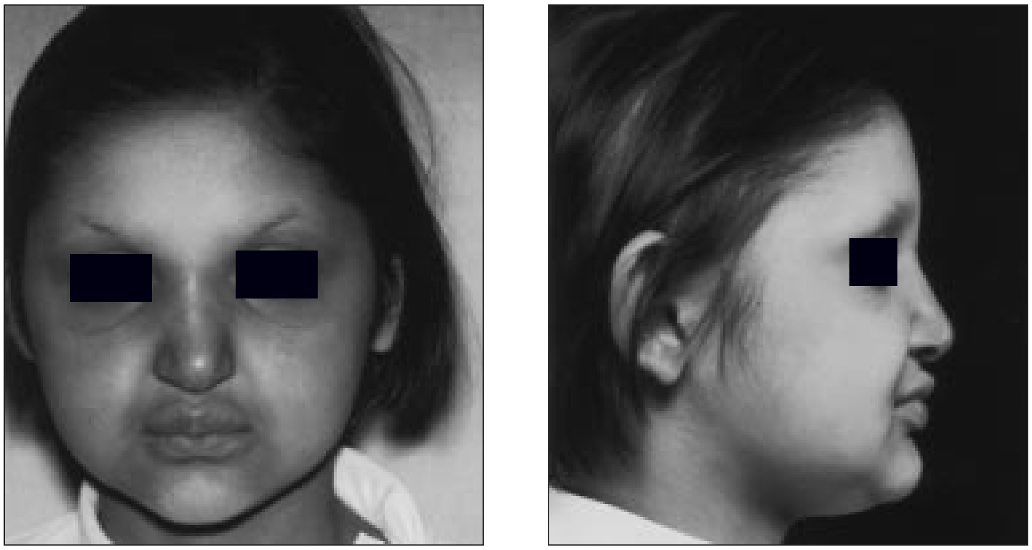
Fig 5c Mandibular superimposition on stable structures and endosseous implants during the same period.

followed the downward and forward displacement of the maxilla and mandible (9.1 mm forward in the maxilla, 17.9 mm in the mandible; Fig 5a). The vertical displacement of the maxilla was only 2 mm, and the relocation of the nasal floor and maxillary sinus floor was 3.8 mm, slightly more pronounced anterior than posterior (Fig 5b), which was the result of the forward (counterclockwise) rotation (2.5 degrees) of the maxilla. The mandible rotated forward (counterclockwise) 10.5 degrees (Fig 5c), which caused a change in the inclination of the implants (70 degrees to 68.5 degrees; Fig 5a). Bone apposition could be observed inferior to the symphysis (Fig 5c). No transverse enlargement could be measured posteriorly on models from age 9 to 20 years.

Figures 6a to 6d show the lateral cephalometric radiographs from ages 8 to 20 years, and Figs 7a to 7d show the patient at ages 8 and 20 years.



Figs 6a to 6d Lateral cephalometric radiographs at (a) age 8 years, (b) age 9 years, (c) age 19 years, and (d) age 20 years. Note relocation of the nasal floor from 9 to 19 years and the eruption of the mandibular first molars.



Figs 7a and 7b Patient at first visit at age 8 years.

Figs 7c and 7d Patient at age 20 years, 12 years after initial treatment.

Table 2 Reported Mandibular Endosseous Implants in Growing Patients with Ectodermal Dysplasia

Authors	Subjects (no./sex)	Age (y)	Mandibular dentition	Implants (no./length)	Region	Follow-up (y)	Growth analysis	Other
Bergendal et al[20] (1991)	1/male	6	Edentulous	2/10 mm and 2/13 mm Brånemark	Between mental foramina	4	No	Significant bone growth around implants
Escobar and Epker[21] (1998)	2/male	7 and 11	Edentulous (both patients)	5 Brånemark (both patients)	Between mental foramina (both patients)	5 to 6	No	Significant bone growth around implants
Cronin et al[16] (1994)	2/female	5	Partially edentulous	2	First incisor region	None	No	
		14	Partially edentulous	6	Right to left second pre-molar region	None	No	Several mandibular teeth extracted
Kearns et al[17] (1999)	6/male	11.2 (mean)	4 patients edentulous, 2 patients partially edentulous	22 (total) 3i and Brånemark	Between mental foramina	7.8 (mean)	Implants follow forward growth of mandible	Two patients had bone grafts; 1 implant did not osseo-integrate
Davarpanah et al[22] (1997)	1/male	14	Partially edentulous	4/10 mm Brånemark	Between mental foramina	None	No	
Smith et al[23] (1993)	1/male	5	Partially edentulous	1/13 mm Brånemark	Central in the anterior mandible	None (done in later study)	No (done in Kearns et al[17]); submerged implant	
Guckes et al[24] (1997)	1/male	3	Edentulous	4 IMZ	Between mental foramina	5	Implants follow forward growth of mandible No angulation problems	

Brånemark: Nobel Biocare, Göteborg, Sweden; 3i: Implant Innovations, Palm Beach Gardens, FL; IMZ: Interpore International, Irvine, CA.

DISCUSSION

The use of endosseous implants in the oral rehabilitation of adolescent patients with HED should be considered a viable treatment option. Opinions vary as to whether it is advisable to place endosseous implants in growing patients, since there is a lack of scientific knowledge concerning the fate of these implants and associated prosthetic rehabilitation. Also unknown is what happens psychologically to these patients when no treatment or various temporary solutions are provided.

The present female patient, who was treated before puberty with endosseous implants and followed for 12 years, developed well psychologically. The success achieved in this patient suggests it is possible to treat patients before puberty with endosseous implants. Only 1 additional surgical procedure and 1 prosthesis remake were required during the 12-year follow-up.

Endosseous Implants in Partially Edentulous Developing Jaws

Considerable research supports the efficacy of rehabilitating a completely or partially edentulous mandible and maxilla using prostheses supported by implants.[4,5] However, almost all the scientific investigations have been performed in adults, when the dynamics of growth and development are not an issue. Placement of implants in the growing maxilla and/or mandible that is only missing a few permanent teeth has been studied, and it has been demonstrated both clinically[6–8] and experimentally[9,10] that ankylosed (osseointegrated) endosseous implants adjacent to the natural teeth become submerged because of the continued eruption of the neighboring teeth and associated growth of the alveolus. A majority of studies have therefore advocated delaying implant placement until skeletal growth is completed, especially when natural teeth are present.[6,11]

Table 3 Reported Maxillary Endosseous Implants in Growing Patients with Ectodermal Dysplasia

Authors	Subjects (no./sex)	Age (y)	Maxillary dentition	Implants (no./type)	Region	Follow-up (y)	Growth analysis	Other
Kearns et al[17] (1999)	6/male	14 12 12 7	Edentulous Edentulous Partially edentulous Partially edentulous	Altogether, 19 3i and Brånemark	In the anterior maxilla (edentulous subjects); in the buccal segments (partially edentulous subjects)	7.7 (mean)	No No No No	Three oldest patients had bone grafts; submerged implant in the 7-year-old
Guckes et al[24] (1997)	1/male	3	Left and right canine present	2 IMZ	Anterior maxilla	5	No	Both implants submerged

3i: Implant Innovations, Palm Beach Gardens, FL; Brånemark: Nobel Biocare, Göteborg, Sweden; IMZ: Interpore International, Irvine, CA.

Endosseous Implants in Edentulous or Nearly Edentulous Developing Jaws

Some studies and case reports have described the placement and follow-up of mandibular endosseous implants in patients with HED (Table 2), but limited knowledge is available concerning implant placement in the edentulous or nearly edentulous growing maxilla (Table 3). Before placing dental implants in the growing mandible or maxilla, the clinician should have an understanding of the growth and development in this region.

Mandibular Growth Considerations and Endosseous Implants

From a developmental perspective, the mandible consists of basal bone (body, ramus, and symphysis) and alveolar bone. Three processes are also present: condylar, coronoid, and angulus. Björk and Skieller[12] showed in a group of healthy untreated adolescents that the average forward rotation was 6 degrees during the 6 years around puberty. The influence of edentulism on mandibular growth is not well understood. In the congenital edentulous mandible, it is mainly the alveolar process that shows pathologic development, because of the lack of erupting teeth. It has been shown that subjects with multiple congenital missing teeth, on average, have forward rotation of the mandible[13] and a reduced mandibular plane angle.[14]

In the absence of the restoration of normal function, it is likely that the expression of facial growth would have been severely affected by the edentulism seen in this patient. Normal function helps build and maintain bone mass. Suboptimally loaded bones atrophy by increasing the remodeling frequency while inhibiting osteoblast function.[15]

In the analysis of the present subject, normal growth of the condyle was demonstrated (Figs 5a and 5c). The cephalometric tracing shows continued condylar growth, whereby the mandible and the implants are displaced forward. This finding is consistent with the findings in the studies mentioned in Table 2. The mandible in the patient under consideration rotated 10.5 degrees forward (4.5 degrees more than average). The implants were not submerged because of lack of vertical growth of the alveolar process in the anterior part of the mandible. It was interesting to see the actual vertical growth in the molar region, which became more pronounced because of the forward rotation (Fig 5c).

During mandibular rotation the teeth normally adapt to the rotation by compensation in eruption and inclination,[12] but because of the osseointegration process, this is not the case for dental implants. The changed inclination of the implants caused by the mandibular rotation can be observed on the superimposition on the anterior cranial base (Fig 5a and Table 1). The possible problem of changed inclination of the implants related to mandibular rotation has been discussed,[16] but no documentation was presented. In the studies shown in Table 2, the possible change in inclination was mentioned in only one study,[17] and the authors reported that inclination of the implants was unchanged; however, the study did not describe the mandibular rotation pattern of the 6 subjects. Continued vertical eruption of natural teeth and the rotation pattern of the mandible therefore seem to be the limiting factors for placing implants in the growing mandible.

In the edentulous or nearly edentulous mandible, as in the presented subject, where the present teeth were not in proximity with the implants, the only change in the implant position would be the inclination. This can be compensated for by revising the prosthesis.

The surface remodeling of the mandibular plane normally associated with mandibular rotation[18] did not have a negative influence on the implants, since apposition occurred inferior to the symphysis and resorption took place at the angulus border (Fig 5c). This is the typical remodeling pattern of forward rotating mandibles.[18] Transverse growth in the anterior region ceases early (before 1 year of age)[18] and is therefore not influenced.

Maxillary Growth Considerations and Endosseous Implants

Maxillary growth is much more complex than mandibular growth, and the end result of an osseointegrated implant placed during growth is difficult to predict. Maxillary growth occurs as a result of both passive displacement and enlargement.[18,19] Passive displacement occurs as the maxilla is carried downward and forward by growth and flexion of the cranial base and a complicated system of sutures in the midface.[18] Maxillary enlargement occurs in vertical, sagittal, and transversal dimensions.

- *Vertical Enlargement.* The increase in maxillary height takes place by growth in the alveolar process in association with dental eruption. In connection with the vertical growth, the hard palate is remodeled, the upper surface is resorbed, and apposition takes place on the lower aspect of the hard palate and alveolar process. This remodeling enables additional enlargement of the nasal cavity, which is not possible by inferior repositioning of the maxilla alone.[18] Implant studies[18] indicate that this relocation increases when the sutural lowering is diminished.
- *Sagittal Enlargement.* Growth in the length of the maxilla is known to occur suturally toward the palatine bone and by apposition on the maxillary tuberosities. From early infancy, all sagittal enlargement originates from the posterior part of the maxilla.[18]
- *Transversal Enlargement.* Growth in the midpalate suture is the most important factor in transversal enlargement. The posterior part of the maxilla enlarges 3½ times as much as the anterior part.[19] Rigid connection over the midline should therefore be avoided.

In a recent study,[8] the use of implants in the growing maxilla was discouraged. However, a few reports concerning endosseous implants in the growing edentulous or nearly edentulous maxilla can be found in the common databases (Table 3).

In the 2 subjects (Table 3) in whom the implants submerged,[17,24] the implants were placed in the vicinity of natural teeth, and the subjects were only age 3 and 7 years, respectively. In the remaining 3 subjects[17] and in the present subject, severe impaction was not encountered. This is the result of the relative lack of vertical growth of the edentulous alveolar process. The amount of vertical growth that is associated with tooth eruption can be observed in connection with eruption of the canines (Fig 5b).

Severe impaction of osseointegrated dental implants in the maxillary alveolus in connection with remodeling of the nasal floor and hard palate has been pointed out by Cronin and Oesterle.[8] It was postulated that implants in the alveolus of a young, growing maxilla may become significantly buried in bone and their apical portions exposed as the nasal floor and maxillary sinuses remodel. The effect of remodeling in the presented subject can be seen on the maxillary superimposition (Fig 5b). Björk and Skieller[19] showed an average nasal floor remodeling of 4.6 mm in boys aged 4 to 20 years. The inferior repositioning related to surface remodeling in the present patient was 3.8 mm.

Because the continuous lengthening of the maxilla occurred posterior to the implants, the implants moved in harmony with the sagittal displacement and growth of the maxilla (Fig 5a). No transverse enlargement could be registered in the tuberosity region of models from age 9 to 20 years. By not rigidly connecting the right and left implants, interference at the midline growth suture could possibly be avoided.[17]

CONCLUSIONS

In growing individuals, edentulism can become a functional or cosmetically disabling condition. The literature and this case report suggest that endosseous implants can be placed with a good prognosis in the edentulous or nearly edentulous growing maxilla or mandible, provided the mandibular implants are placed anterior to the mental foramen.

In the mandible, only the rotation had an effect on the implants by changing their angulation. To the authors' knowledge, submerged implants have not been reported in the edentulous mandible, provided they are not placed close to natural teeth. In the growing edentulous maxilla, a certain amount of impaction must be expected because of relocation of the nasal floor and the maxillary sinuses. The amount of relocation can be evaluated only by maxillary superimposition, as demonstrated in this investigation.

Variation in growth from individual to individual and the difficulties in predicting the amount and direction of growth should be considered, but caution must be exercised in generalizing the results. A multidisciplinary approach for oral and maxillofacial rehabilitation of these patients is recommended.

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