Dental Implants in Patients with Ehlers-Danlos Syndrome: A Case Series Study

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Ehlers-Danlos syndrome (EDS) encompasses six types of hereditary connective tissue disorders, with skin hyperextensibility, joint hypermobility, and connective tissue fragility as the main findings. Oral health is also affected, sometimes including periodontitis and tooth loss. This is the first report on dental implant treatment for patients with hypermobility or classic EDS. Five female patients aged 19 to 68 years who tolerated treatment under local anesthesia and did not require bone augmentation were enrolled in the study and received 16 implants. They were observed for 2 to 12 years. No implants were lost, bone loss was minimal, and all patients were pleased with the treatment outcomes. *Int J Prosthodont 2012;25:60–62.*

The Villefranche classification[1] describes Ehlers-Danlos syndrome (EDS) as encompassing any of six types of inherited connective tissue disorders that involve a unique defect in collagen metabolism.[2] EDS is broadly characterized by the clinical signs of skin hyperextensibility, delayed wound healing with atrophic scarring, joint hypermobility, easy bruising, and generalized connective tissue fragility. The prevalence of EDS is between 1 in 5,000 and 1 in 10,000 and is observed throughout the world affecting both sexes. The most common form of EDS, the classic type, is caused by abnormal type V collagen, while the cause of EDS with hypermobility is still unknown. Both types are characterized by skin laxity and joint hypermobility, with the former being more pronounced in the classic type and hypermobility in the latter. Many aspects of oral health are influenced by EDS.[3] Early-onset generalized periodontitis and tooth loss have both been reported.[4] Hence, patients with EDS may require more prosthodontic treatment including dental implants than the general population, although this is unreported. In this paper, preliminary dental implant treatment outcomes of five female patients with EDS are described.

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Table 1 Inclusion and Exclusion Criteria

Inclusion criteria
EDS classic or hypermobility type
Electronic chart and orthopantomogram available
Age > 18 y
Lack of one or more teeth suitable for dental implant treatment without bone augmentation or having implants placed previously
Tolerate treatment under local anesthetics
Exclusion criteria
Additional medical diagnoses putting the patient at surgical risk
Severe pain/dislocation of the temporomandibular joint
Requiring general anesthesia for dental treatment

Materials and Methods

Six patients were recruited from the 119 patients with a reported EDS diagnosis at the National Resource Centre for Oral Health in Rare Medical Conditions, Lovisenberg Diakonale Hospital, Oslo, Norway, according to strict inclusion and exclusion criteria (Table 1). Three patients had already undergone treatment with implants, and 3 additional patients were invited for a consultation. One patient never showed up; the 2 others were treated with dental implants and followed prospectively. A total of 5 women participated in this study. Their demographics are given in Table 2. The Regional Medical Ethical Committee, East, Norway, approved the study (610-07-062461.2006-2244), and informed consent was obtained from each patient.

Surgical and Prosthodontic Procedures

The surgical procedures for all patients but one were performed at the University of Oslo using Astra implants (Astra Tech); patient 1 was treated with a

Table 2 Patient Demographics

Patient	Year born	EDS type	Smoker	Symptoms of EDS	Work status	Medication
1	1979	H	No	Skin,‡ hypermobile, joint pain	Working part-time due to EDS	Antiphlogistics, analgesics
2	1936	H	No	Skin,‡ hypermobile, joint pain	Retired, quit working due to EDS	Antihypertensives, analgesics, antiphlogistics
3	1980	H	Yes	Hypermobile, joint pain, skin‡ (hernia)	Not working due to EDS	Antiasthmatics, analgesics, antiphlogistics
4	1964	C*	Yes†	Skin,‡ hypermobile, joint pain, circulatory insults	Not working due to EDS	Anticoagulants, antihypertensives, thyroid hormones, lipid-modifying drugs
5	1959	H	No	Skin,‡ hypermobile	Working part-time due to EDS	Antiphlogistics, antihistamines, analgesics

C = classic; H = hypermobile.
*Reclassified from H after surgery.
†Stopped smoking September 2008 (after surgery).
‡Skin laxity, delayed wound healing, scarring, or hematomas.

Table 3 Implant Characteristics and Observation Times

Patient	Age at implant insertion (y)	Implant site*	Date of implant placement (brand)	Implant dimensions (mm)	Date of abutment placement	Date of prosthetic loading	Date of last control	Prosthetics
1	19	14	Nov 1998 (Brånemark Mark 2)	3.75 × 13	Dec 1999	May 2000	Nov 2010	Single cemented crown
2	68	11 21 23	Aug 2004 (Astra TiOblast)	3.5 × 13 3.5 × 13 3.5 × 13	Feb 2005	April 2005	Nov 2010	Six-unit screw-retained FPD; new FPD because of porcelain fractures in Dec 2008
		35 44 45	Oct 2004 (Astra TiOblast)	3.5 × 11 3.5 × 11 3.5 × 11				Single screw-retained crowns
3	26	25	June 2006 (Astra Osseospeed)	3.5 × 11	June 2006 (single-stage)	Oct 2006	Nov 2010	Single cemented crown
4	43	14 12 11 21 22 23	Nov 2007 (Astra Osseospeed)	4.0 × 13 4.0 × 11 4.0 × 11 4.0 × 11 4.0 × 11 4.0 × 11	May 2008	Sept 2008	Dec 2010	Ten-unit screw-retained FPD
5	49	36 46	Nov 2008 (Astra Osseospeed)	4.0 × 11 4.0 × 9	March 2009	May 2009	Nov 2010	Single screw-retained crowns

FPD = fixed partial denture.
*FDI tooth-numbering system.

Brånemark implant (Nobel Biocare) at the University of Bergen. Two-stage procedures were performed for all but one implant. Prosthodontic procedures were performed by registered prosthodontic specialists.

Evaluation of Implants

Implant survival was recorded. Bone loss after loading was registered as the mean of the mesial and distal bone levels of each implant to the closest millimeter, as judged on periapical radiographs using the parallel technique and film holders. Gingival status around implants was scored as follows: 0 = ideal, 1 = mild color change, 2 = moderate bleeding on probing, and 3 = spontaneous bleeding. At the final follow-up, the treatment outcome was evaluated by both patients and specialists regarding esthetics, speech, and function, ranging from 0 (worst) to 10 (best).

Results

The patients had many symptoms related to EDS, were taking analgesics and additional drugs, and were working part-time or not at all as a result of EDS.

Table 4 Patient Satisfaction with Dental Implants*

Patient	Overall satisfaction	Esthetics	Speech	Function
1	10	10	10	10
2	9	10	10	10
3	9	10	10	9
4	10	10	10	10
5	10	10	10	10
Mean	9.6	10.0	10.0	9.8

*Visual analog scale from 0 (worst) to 10 (best).

Table 5 Evaluation of Gingival Status Around Implants, Esthetics, Speech, and Function by the Specialist Team

Patient	Implant site*	Gingival Index†	Esthetics‡	Speech‡	Function‡
1	14	1	10	10	10
2	11	1	8	10	10
	21	1	8		
	23	1	8		
	35	1	5		
	44	1	3		
	45	1	3		
3	25	2	8	10	10
4	14	2	10	10	10
	12	2	10		
	11	1	10		
	21	2	10		
	22	1	10		
	23	2	10		
5	36	1	9	10	10
	46	1	9		
Mean		1.3	8.2	10.0	10.0

*FDI tooth-numbering system.
†Scale from 0 to 3, see text for details.
‡Visual analog scale from 0 (worst) to 10 (best).

Each patient was missing 5 to 18 teeth. A total of 16 implants were placed in the five patients. The mean age at implant insertion was 41 years (range: 19 to 68 years). Implant characteristics and observation times are given in Table 3. The patients were followed for a mean period of 5.5 years after insertion (range: 2 to 12 years). No implants were lost. At the last follow-up, all prosthetic reconstructions were in place. No significant bone loss was seen in three patients. Patient 4 had bone loss of 2 mm around one implant in the maxillary anterior region, and patient 2 had bone loss of 2.7 mm around an implant in the mandibular premolar region. The results of the subjective and objective evaluations of the implants are given in Tables 4 and 5, respectively.

Discussion

This is the first study on implants in patients with EDS. A high success rate was demonstrated employing the strict inclusion criteria. Because of practical and ethical considerations as well as the small size of most patient groups with various rare disorders, the possibilities for designing prospective studies yielding a high level of scientific evidence are limited.[5] Patients with EDS are a heterogenous group, including smokers and nonsmokers, with varying degrees of treatment complexity and medical conditions.

Conclusion

Within the limitations of the study design, it is suggested that dental implants can be placed in patients with EDS with almost the same success rates as in healthy individuals as well as to the satisfaction of the patients.

Addendum

This paper was submitted in March 2011. Since then, patient 4, who received six maxillary implants in 2007, had to have most of her mandibular teeth removed because of severe periodontitis. She did not tolerate a partial mandibular prosthesis, and four mandibular implants (Astra Osseospeed) were placed in October 2011. Unfortunately, this was the patient with the most advanced periodontal disease in addition to, or as part of, EDS. However, the initial bone loss observed around one of the maxillary implants has not progressed during 2011. Hopefully the prognosis of the implants will be better than that of her own dentition. The reader's attention is drawn to the recent relevant commentary by Birgitta Bergendal in the number 6, November 2011, issue of the IJP. She noted 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."

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