Title page

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Dental implants in individuals with osteogenesis imperfecta: a 6-year follow-up study

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Abstract

Dental implants in individuals with osteogenesis imperfecta: a 6-year follow-up study

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Abstract

Aim: To perform a long-term follow-up of our previous prospective study, which after a mean observation time of 1.5 years reported high implant survival rate in a group of individuals with osteogenesis imperfecta (OI). Our hypothesis was that long-term implant survival rate in these individuals is approximately the same as in healthy individuals.

Study group: The previous study included seven participants (20 implants), four of them (11 implants) took part in the present study, the other three had died. The participants were followed-up for an average of 93 months, subsequent to prosthetic loading. The implants were clinically and radiographically examined. Objective and subjective evaluations were recorded using an analogue scale ranging from 0 as the worst to 10 as the best score. A mean of these evaluations is presented as the subjective and objective overall satisfaction.

In the previous study, no implants were lost, and 1 mm bone loss was recorded around two implants. One implant was removed after 76 months due to an implant-neck fracture. At the present study, four implants showed 1 mm bone loss, two of which had the same level of bone loss in the previous study. 4 mm bone loss was observed around two other implants. No bone loss was detected around the remaining four implants. Objective and subjective evaluation of implant treatment, showed an overall high satisfaction of 9.1 and 9.9, respectively.

Conclusion: The findings showed an implant survival rate of 91% (100 %, excluding the implant-neck fracture) and high recipient satisfaction towards implant treatment.

Clinical relevance

Scientific rationale for study

This long-term study is based on our previous short term study conducted in 2011.

Principle findings

Eleven implants in four patients were followed up for six-years. No bone loss was observed around four implants, 1 mm bone loss around four implants, and 4 mm bone loss around two implants. One implant was removed due to an implant-neck fracture.

Practical implications

The results show that long-term outcome of dental implant treatment in individuals with OI in this study had a high implant survival rate.

Introduction

Osteogenesis imperfecta (OI) is the most common inherited form of bone fragility and includes a heterogeneous group of disorders which most commonly result from defects associated with type 1 collagen. Most cases (85%–90%) are inherited in an autosomal dominant manner and are caused by mutations in the collagen COL1A1 or collagen COL1A2 genes, leading to quantitative and/or qualitative defects in type 1 collagen¹. In addition to the skeletal phenotype, common additional extraskeletal manifestations include blue sclerae, vascular fragility, hearing loss, and dentinogenesis imperfecta (DI).

Four major types of OI (I, II, III and IV) based on clinical features, severity and mode of inheritance were originally described². Type I is a mild form of OI. Type II is the lethal form of OI, even during the prenatal and perinatal period. Individuals with type III of OI show progressive limb deformation, whereas in type IV moderate to severe phenotypes are seen. OI types V-VIII were added later because of distinct clinical features and/or different causative gene mutations³. Clinical and radiological features of OI type VII and VIII are indistinguishable from OI types II-IV, but were added because of genetic criteria (autosomal recessive inheritance)⁴.

Dentinogenesis imperfecta (DI) is a common feature in OI⁵. Depending on the type of OI, prevalence of DI is between 8% and 100%^{6, 7}. Andersson *et al.*⁸ recently reported that the prevalence of DI was highest in children with OI type III and lowest in children with OI type I (86% vs. 31%). Many individuals with OI also present other dental aberrations including tooth agenesis (congenital absence of one or more permanent teeth). Malmgren *et al.* reported that the prevalence of tooth agenesis is high (17%) in individuals with OI⁶, and OI caused by a qualitative collagen I mutation is associated with oligodontia⁸.

Dental implants have been safely installed to replace missing teeth for half a century and proved to be a safe procedure with reported survival rates exceeding 95%^{9, 10}. Although initially installed only in healthy individuals, dental implants have been increasingly placed in medically compromised persons since evidence on the effect of systemic diseases on dental implant success is limited¹¹. A few case reports have previously described successful implant treatment in persons with OI^{12, 13, 14, 15, 16, 17}, whereas no study on the survival rate of dental implants in a group of individuals with OI had been published until our study in 2011¹⁸.

The aim of the present study was to follow up our previous prospective study that reported high implant survival in a group of Norwegian individuals with OI. Our hypothesis was that implant treatment in these persons has approximately the same long-term survival rate as in healthy individuals.

Methods and participants

The previous approval from the Regional Medical Ethical Committee, East, Norway, (S2006-2244) was renewed for this follow-up study.

The seven individuals who participated in the previous study were all previously examined at the National Resource Centre for Oral Health in Rare Diseases (TAKO-center), Lovisenberg Diaconal Hospital, Oslo,

Norway and were now invited to take part in the 6-year follow-up study. Among them, three individuals had died, and the remaining four individuals agreed to participate in the present study. They all have a diagnosis of OI and received dental implants due to agenesis or loss of teeth (Table 1). The participants received written information. After having obtained signed consents, a clinical examination was carried out and an orthopantomogram and periapical radiographs were taken.

Age, gender, type of OI and previous treatment were reconfirmed. The subgroups of OI were classified according to Sillence clinical classification (I-IV)¹⁹ with 'b' indicating DI. One of the participants (ID 7) used bisphosphonates for a period after completion of the previous prospective study. She developed peri-implantitis thereafter. The crown was removed, and a surgical procedure was performed to clean the implant surface. After four months, bone had successfully regenerated around the implant and the crown was remounted.

Surgical and prosthodontic procedures

Detailed surgical and prosthodontic procedures were described in our previous study¹⁸.

In short, each participant received one, two, three, or five implant/s respectively. Two-stage surgical procedures for three participants and a one-stage procedure for one participant were performed between 2007 and 2009 at the Department of Oral Surgery and Oral Medicine, University of Oslo. Prosthodontic procedures were performed by registered prosthodontic specialists.

Evaluation of implants

Two dentists, one of whom examined the participants in the previous study, evaluated the peri-implant bone levels by judging periapical radiographs taken with the parallel technique using film holders. Bone loss was visually assessed at periapical radiographs at the follow-up study and compared to that at loading. Implant characteristics, observation times, and peri-implant bone loss are presented in Table 2.

Gingival status around the implants was scored as: 0: ideal, 1: mild color change, 2: moderate bleeding on probing, and 3: spontaneous bleeding. The team's objective evaluations of aesthetics, speech, and function were recorded (Table 3). In addition, the participants' subjective opinions on aesthetics, speech, and function were registered (Table 4). Means of these evaluations are presented as the objective and subjective overall satisfaction, respectively.

Results

The characteristics of the participants are presented in Table 1. The mean age in years at the time of the present study was 56.3. The implants and the status of peri-implant periodontium were initially examined after an average of 19 months (range 11-26 months) and followed up after an average of 93 months (range 91-109 months), subsequent to prosthetic loading.

At the previous study, no implants were lost, and only 1 mm bone loss was registered around two implants in one participant. One implant was removed after 76 months due to an implant-neck fracture, unrelated to disease. At the follow-up study, 4 mm bone loss around two implants was observed in two participants. Four implants showed only 1 mm bone loss, two of which had the same level of bone loss at the previous study. No bone loss was detected around the remaining four implants. The observation of peri-implant bone loss is presented in Table 2 and Fig. 1.

Evaluation of peri-implant gingiva showed moderate bleeding on probing around six implants, whereas spontaneous bleeding was not observed. Mild translucency of metal color through gingiva was seen around 7 implants.

The objective evaluation of aesthetics showed a mean score of 7.3 with a range of 5-10, while speech was scored as 10 for all implants. With regard to function, nine implants were scored as 10 while the remaining were scored as 9. The participants' subjective opinions on aesthetics showed a mean score of 9.8 with a range of 9-10, while speech and function were scored as 10 for all implants. The objective and subjective evaluation of implant treatment, respectively, showed overall satisfaction of 9.1 and 9.9 after the follow-up study. The results of the evaluations are given in Tables 3 and 4.

Discussion

Our long-term follow-up study clearly demonstrated a high survival rate of bone-anchored prosthetic restorations in individuals with OI in this study.

This is the first study reporting long-term follow-up in a group of individuals with OI treated with dental implants. Four out of seven individuals who participated in the previous study took part in the present follow-up study. The remaining three passed away during the course of 6 years, due to other reasons not related to implant treatment. Two of the deceased individuals had OI type I and had a similar length of life as the general population²⁰, while the one with OI type III had a shorter life. We have not received any information indicating implant loss in these three individuals.

Among the four participants in the follow-up study, two were smokers and one received bisphosphonates for a short time. One of the two implants that showed 4 mm bone loss was placed in a smoker and the other in a non-smoker. The other smoker, who used bisphosphonates for a period after implant treatment developed peri-implantitis that was successfully treated. These observations indicate that many factors in addition to OI are to be considered.

In the previous study¹, no significant peri-implant bone loss was observed after an average of 19 months subsequent to prosthetic loading, and the survival rate of the 20 implants was 100%. The present study, in which 11 implants from the previous study were followed up for 93 months, showed 4 mm bone loss around two implants. Additionally, one implant was lost 76 months after installation due to a fracture of the implant itself. Therefore, none of the implants were lost due to peri-implant pathology. Thus, total survival rate of the implants still stands at 100% or at 91% when counting the implant-neck fracture. High implant survival rate observed in this study is probably influenced by determining factors such as type of OI, quantity/quality of bone, appropriate choice of implants, location of implant placement, oral hygiene maintenance and underlying medical condition of the participants.

OI type I is the most common and usually the mildest form of OI. Lund *et al.*²¹ reported that OI type I individuals with DI (OI type Ib) were more severely affected by OI than those without DI. Two of the four participants in the present study have OI type I with DI, and in these individuals only one implant out of eight had 4 mm bone loss. Individuals with OI type Ib in this study do not clearly demonstrate a poorer prognosis regarding implant treatment than OI type I individuals without DI.

In the present study, we were only able to follow-up four out of seven individuals with OI as three of them died. We are aware that too small a sample may not be representative of the overall OI.

In conclusion, the results of our 6-year follow-up study demonstrates that dental implants placed in individuals with OI in this study had almost the same long-term survival rate as in healthy individuals, and that recipient satisfaction towards implant treatment was high. Further research on dental implant treatment in larger groups of individuals with different types of OI, accompanied by control groups is warranted to determine if our findings are indicative of general OI population.

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Table 1. Participant characteristics

ID*	Year Born	Gender	Type of OI	Smoker
3	1953	M	Ib**	Y
5	1972	M	Ib	N
6	1963	F	I	N
7	1955	F	IV	Y

*Three persons had died during the follow-up period; the ID numbers are kept as a reference to the previous study

Ib**, OI type I with dentinogenesis imperfecta

Table 2. Implant characteristics, observation times and peri-implant bone loss

ID	Implant site	Brand	Implant diameter	Implant length	Previous study		Follow-up study	
					Months after installation	Bone Loss (mm)	Months after installation	Bone Loss (mm)
3	24	AS	4	13	26	0	109	4 ^d
	25	AS	4	13	26	1 ^b	109	1 ^c
	45	AS	4	13	20	0	103	1 ^c
	46	AS	4	13	20	1 ^b	103	1 ^c
	47	AS	4	11	20	0	103	0 ^e
5	25	AS	3,5	13	22	0	106	0 ^e
	31	3I	3,25	11,5	20	0	104	0 ^e
	41	3I	3,25	13	20	0	104	1 ^c
6	15	AS	5	11	11	0	91	4 ^d
	36 ^a	AS	4,5	11				
7	36	S	4,1	8	11	0	94	0 ^e

3I = 3I tapered; AS = Astra Ossepeed; S = Straumann

^a: The implant was removed after 76 months due to an implant-neck fracture, unrelated to disease.

^b: 1 mm peri-implant bone loss (previous study)

^c: 1 mm peri-implant bone loss (follow-up study)

^d: 4 mm peri-implant bone loss (follow-up study)

^e: No peri-implant bone loss (follow-up study)

The corresponding peri-implant bone loss or lack of it are shown at the radiographic images in Figure 1.

Table 3. Objective evaluation, visual analogue scale 0-10 (0 as the worst and 10 as the best)

ID	Implant site	Aesthetics	Speech	Function	Overall satisfaction
3	24	7	10	10	
	25	10	10	10	
	45	7	10	10	
	46	7	10	10	
	47	10	10	10	
5	25	5	10	9	
	31	5	10	10	
	41	5	10	10	
6	15	8	10	10	
	36*	-	-	-	
7	36	9	10	10	
Total		7.3	10	9.9	9.1

*The implant was removed after 76 months due to an implant neck fracture, unrelated to disease.

Table 4. Subjective evaluation, visual analogue scale 0-10 (0 as the worst and 10 as the best)

ID	Aesthetics	Speech	Function	Overall satisfaction
3	10	10	10	10
5	9	10	10	9.7
6	10	10	10	10
7	10	10	10	10
Total	9.8	10	10	9.9

Figure 1. Clinical and radiographic examination of the implants

- b: 1 mm peri-implant bone loss (previous study)
- c: 1 mm peri-implant bone loss (follow-up study)
- d: 4 mm peri-implant bone loss (follow-up study)
- e: No peri-implant bone loss