Clinical Considerations of Ehlers-Danlos Syndrome for Implant Dentistry: Two Case Reports

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Ehlers-Danlos Syndrome (EDS) is a group of congenital connective tissue disorders that commonly affect joints, muscles, soft tissue, and blood circulation in the affected population. Many oral manifestations are displayed in EDS patients, including gingival recession, lack of attached gingiva, early severe periodontal disease, and dental caries. However, the literature is limited and oftentimes contradictory regarding dental implants in EDS patients. The aim of this study is to report two successful cases of dental implants placed in EDS patients, one treated with bone augmentation and both restored with prosthetic implant rehabilitations. *Int J Periodontics Restorative Dent* 2022. doi: 10.11607/prd.5842

### INTRODUCTION

Connective tissue disorders (CTDs) are a heterogeneous group of diseases that commonly affect the connective tissue layers of the body. The function of these tissues is predominately to bond one or more structures of the body including skin, bone, blood vessels and muscles (1). In the affected population, collagen and elastin are inflamed and oftentimes defective, leading to various complications including tissue fragility with limited strength and elasticity (2).

One of the most commonly reported CTDs is Ehlers Danlos Syndrome (EDS) with a prevalence of 1/1500 in births (3). The etiology of EDS is mainly related to autosomal dominant or recessive genetic disorders, which are inherited and passed on from parent to child. As of today, there is no treatment for EDS. Pathognomonic features of EDS are joint hyperextensibility, scarring, decreased wound healing and easy bruising (4). Due to this collagen disorder, many oral manifestations are displayed in EDS patients. The most common are enamel hypoplasia, deep fissures in the premolars and molars leading to dental caries, calcification of the pulp chambers, thin or lack of attached gingiva, gingival recession,
and early severe periodontitis (5-7). One or all of these symptoms may be present in a patient with EDS. In regard to dental implant therapy in EDS patients, the literature is still limited and controversial due to the poor healing capacity in patients suffering EDS (8,9). Furthermore, a search of the literature reveals a lack of evidence-based data concerning implant dentistry, bone grafting, sinus augmentation and other surgical regenerative techniques.

Therefore, the purpose of this study is to present two case reports of EDS patients that were successfully treated with dental implants which required augmentation procedures.

MATERIALS AND METHODS:

CASE 1:

A 63-year-old female patient was referred to the Ashman Department of Periodontics and Implant Dentistry of New York University College of Dentistry (NYUCD) for evaluation of multiple missing teeth. The patient was diagnosed with congenital Ehlers Danlos disease type IV and she had bilateral aneurysm of her carotid arteries which were previously treated surgically. She was taking aspirin 81 mg, atorvastatin 40 mg, and vitamins supplements on daily basis. The patient denied use of tobacco cigarettes, alcohol, or any other recreational drugs. The patient’s chief concern was to have her missing teeth restored with dental implant supported restorations refusing other alternative options including removal partial dentures or a fixed dental prosthesis (FDP). The patient was discouraged as many clinicians refused to treat her due to her complex medical history and she came to New York University College of Dentistry seeking assistance. A clinical evaluation revealed that she was missing the maxillary right first premolar, maxillary left second premolar and first molar and mandibular right second premolar and first molar [Figure 1A, 1B]. A soft and hard tissue examination revealed minimal probing depth and no bone loss around the remaining teeth. However, heavy calculus and plaque deposit and generalized gingival recession on the mandibular arch from tooth #19 to tooth #27 were
present [Figure 2]. After a detailed medical consultation with her primary physician, her endocrinologist, and her cardiologist, the patient was medically cleared for the surgical dentoalveolar procedures and implant placement. Risks and benefits of implant surgeries were explained in detail to the patient including the possibility of a failure of the implants, sinus complications including sinusitis or sinus membrane perforation, and failure of the graft and the possibility of having to repeat the procedure. This information was discussed thoroughly with the patient who understood all the risks and signed an informed consent for the different surgical procedures outlined. The patient wished to start with the maxillary left edentulous site (site #13,14).

A Cone Beam Computed Tomographic (CBCT) scan was taken prior the surgery and exhibited a pneumatized maxillary left sinus that required sinus augmentation to allow implant placement [Figure 3]. Prior to the procedure, the patient was scheduled for several full mouth scaling and root planning sessions and was instructed in proper oral hygiene care. When the soft tissue evaluation showed no probing depth, no bleeding on probing, and the patient’s homecare was excellent, the implant phase of treatment began.

On the day of the surgery, the patient was prescribed an antibiotic (Amoxicillin 2 gm) to take one-hour prior the procedure. The maxillary left edentulous site was anesthetized with local anesthesia (Lidocaine HCL 2% with epinephrine 1:100,000, Henry Schein Inc, Melville, NY). A full thickness mucoperiosteal flap was performed from teeth #12-15 with midcrestal, mesial, and distal vertical releasing incisions. The flap was reflected apically to expose the lateral bony wall of the maxillary sinus. The removal of the lateral window was performed with the use of high speed #8 diamond round bur. After removal of the lateral window, the Schneiderian membrane appeared to be healthy and intact with a proper thickness which allowed careful elevation without any perforation [Figure 4]. Following membrane elevation, a small and large particle xenograft (BioOss; Geistlich Pharma North America, Princeton, NJ) were placed coronal to the elevated membrane and the 6 mm of crestal bone present
allowed two implants (3.3 x 12 mm Bone Level Roxolid Tapered SLActive, Straumann, Switzerland and 4.1 x 12 mm; Bone Level Roxolid SLActive, Straumann, Switzerland) to be placed simultaneously in sites #13, 14 [Figure 5]. The implants were placed with a good primary stability achieving a torque of 35 Ncm. The flap was then advanced and secured with a tension free primary closure with 4/0 chromic gut sutures (Henry Schein, Melville, New York) [Figure 6]. Following the surgery, the patient was prescribed antibiotics (Amoxicillin 500mg TID) an analgesic (Ibuprofen 600 mg every 6h) and 0.12% Chlorhexidine mouth rinses for two weeks. The patient was also placed on a soft diet for two weeks. Two weeks later the patient returned for routine follow up with a healing within normal limits, and no evidence of complications.

One month later, the patient returned for surgical placement of the maxillary right first premolar implant and the mandibular right 2nd premolar and first molar implants. These three implants were placed following the manufacturer’s recommendations with a surgical stent made from an ideal wax-up [Figure 7A,7B]. These areas healed uneventfully, and no complications were observed. All the implants were allowed to integrate for a period of four months. At that time, second stage surgery was performed and different sized platform switching healing abutments were connected to the implants to shape the peri-implant soft tissue and to allow proper emergence profile. Three weeks later polyether fixture level pick-up impressions were taken. Screw-retained monolithic zirconia crowns were then fabricated to restore all the five implants that were placed [Figure 8A-8C]. The patient was very satisfied with the outcome of the procedure. Two years post op a clinical examination and periapical radiograph confirmed stable marginal bone levels around the implants. [Figure 9A-9C]

CASE 2:

A 50-year-old female patient presented to the Ashman Department of Periodontics and Implant Dentistry of New York University College of Dentistry for evaluation of edentulous site #29. The
patient had congenitally missing second premolars and the remaining second deciduous molar presented with mobility and diagnosed as hopeless [Figure 10]. The patient was diagnosed with Ehlers Danlos type IV. As in the previous case medical clearance was obtained with a detailed consultation from her primary care physician. The site was planned for immediate implant placement and a CBCT was taken [Figure 11]. The tooth was extracted as atraumatically as possible and an implant (Nobel Biocare Replace Select Tapered 4.3 x 10mm, Yorba Linda, California, USA) was placed with a surgical guide, following the manufacturer’s protocol [Figure 12]. No intra or postoperative complication were encountered, and the patient’s post-operative course was unremarkable. Three months later, second stage surgery was performed. Following the healing, an open tray impression was taken, and a screw retained porcelain fused to metal (PFM) crown was delivered to the patient [Figure 13,14]. The patient presented for routine follow up every 6 months for 10 years. Periapical x-rays were taken 10-years post implant placement and showed stable levels of peri-implant marginal bone [Figure 15]. The patient was satisfied with the esthetics and function of the crown and no evidence of intraoral inflammation or infection were observed at the 10 year follow up appointment.

DISCUSSION

Implant therapy in EDS patients has, to date, not been thoroughly addressed in the literature. However, four studies are of interest. Jensel et al., in a case series, on the treatment of EDS patients, reported no implant loss and concluded that these patients responded no different than the general population (8). Chapuis et al. findings were in accordance with the previous study with no significant bone loss around the implants placed (10). Borris et al. reported no complication after one year of implant insertion and soft tissue augmentation. (11) Rinner et al., focused solely on three cases of familiar periodontal EDS (type VIII) that were treated with implant therapy. The authors reported severe peri-implant bone loss and advised maintenance of the teeth for as long as possible since
implant prosthetic solutions for these patients carry higher risks of complications (9). In addition, regarding EDS and regenerative techniques including guided bone regeneration (GBR) and sinus augmentation, the current literature is limited. To the author’s knowledge, this is the first clinical case report, that included a sinus augmentation procedure in a patient suffering from EDS. Although connective tissue fragility is inherent in EDS patients, the sinus membrane was found to be within normal limits and the sinus procedure was carried out, with the simultaneous placement of the two implants, without any complications. This case has been restored and in function for more than 2 years without no complications and bone loss. In case 2, the mandibular implant integrated and there was no presence of marginal bone loss or complications after 10 years in function. To prevent complications and failure of the grafted sites, meticulous periodontal maintenance is required in patients suffering from EDS. The patients in these case reports were treated preoperatively to eliminate and control the periodontal problems and instructed on proper home care therapy before and after implant placement. Additionally, neither of the patients treated had deficiencies of attached gingiva and were able to perform adequate daily home care when instructed accordingly. If required, it is advisable to perform soft tissue augmentation prior to the bone regeneration and the implant placement to establish an adequate bond of keratinized tissue.

Based on the success achieved in the cases presented, EDS should not be considered a contra-indication for dental implants and regenerative procedures. Nevertheless, more studies including randomized controlled trials are necessary to validate the results of this study.

CONCLUSIONS

Within the limitation of the current study, it was found that EDS is not an absolute contra-indication for the rehabilitation of partially edentulous patients with dental implants. However, medical consultation with a primary care physician and all necessary periodontal treatment and strict
maintenance protocols were performed prior to and following the implants and sinus augmentation procedure in order to maximize the outcomes. Due to the limited evidence, more studies including randomized controlled trials and systematic reviews and meta-analysis are needed to validate the results of the present study.

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The authors declare no conflicts of interest.

REFERENCES


FIGURE LEGENDS:

Figure 1A: Initial Intraoral Maxillary Occlusal Photograph.
Figure 1B: Initial Intraoral Mandibular Occlusal Photograph.
Figure 2: Initial Intraoral Frontal Photograph displaying generalized deposits of plaque and recession from tooth #19 to #27
Figure 3: CBCT of Maxillary Left Edentolous Site Displaying Pnematization of Maxillary Sinus.
Figure 4: Lateral Window Sinus Augmentation #14.
Figure 5: Simulataneous Sinus Augmentation Technique with Placement of Implant Site #13 and #14.
Figure 6: Primary Closure Tension Free Suture
Figure 7A: Surgical Placement of Implant Site #5
Figure 7B: Surgical Placement of Implant Site #29 and #30

Figure 8A: Final Intraoral Frontal Picture of the patient

Figure 8B: Final Intraoral Maxillary Occlusal Photograph.

Figure 8C: Final Intraoral Mandibular Occlusal Photograph.

Figure 9A: Periapical X-ray of Zirconia Crown of #5

Figure 9B: Periapical X-ray of Zirconia Crowns of #13,14

Figure 9C: Periapical X-ray of Zirconia Crowns of #29,30,31

Figure 10: Preoperative Periapical X-ray of Tooth T with periapical pathology and furcation involvement.

Figure 11: CBCT evaluation of site #28 for immediate implant placement.

Figure 12: Periapical X-ray of Implant Placement Site #29

Figure 13: Intraoral Occlusal Photo of Implant Supported Restoration of #29 at 10 years follow up.

Figure 14: Periapical X-ray of Crown Insertion Site #30

Figure 15: Periapical X-ray of Crown Delivery Site #29 at 10 years follow up.
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Fig 1a

Fig 1b

Fig 2

Fig 3

Fig 4

Fig 5