Prosthetically Driven Therapy for a Patient With Systemic Lupus Erythematosus and Common Variable Immunodeficiency: A Case Report

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Patients who have systemic diseases in conjunction with severely resorbed maxillary and mandibular bone present challenges for dental implant therapy and rehabilitation. This case report describes the interdisciplinary comprehensive treatment completed on a patient with systemic lupus erythematosus (SLE) and common variable immunodeficiency (CVID). Patients with these systemic conditions present a multifactorial challenge for dental treatment due to advanced carious lesions, missing teeth, lack of adequate bone quality and quantity, as well as secondary effects of their medications. The sequence of treatment presented allowed for the necessary case control to ensure successful, predictable reconstruction of the edentulous patient with limited bone available for implant placement. For this patient, we used a combination of autogenous iliac bone graft, bilateral maxillary sinus lifts with BMP-2, transitional implants, and dental endosseous root form implants. Digital dentistry aided in designing the final implant supported fixed restorations. Transitional implants eliminated the need for tissue-borne prostheses, avoiding pressure to the graft and implants. Digital dentistry allowed for prosthetically driven implant placement and a functional, esthetic result. The techniques and staging presented for implant placement and rehabilitation can be used for other patients presenting with similar challenging conditions.

Key Words: lupus erythematosus, systemic, common variable immunodeficiency, dental implants, alveolar ridge augmentation, bone transplantation

INTRODUCTION

Implant therapy for systemically compromised patients with limited bone presents a challenge that requires the expertise of an interdisciplinary team to ensure successful, predictable results. Juvenile-onset systemic lupus erythematosus (JSLE) is one of the most common autoimmune disorders affecting children. The incidence is 0.3 to 0.9 per 100,000 per year worldwide. The disease requires aggressive long-term treatment. Similar to systemic lupus erythematosus (SLE), females are more likely to be affected by JSLE. Clinical presentation tends to be during puberty and is more severe than adult onset SLE, typically with multiple organ systems involved. The diagnosis of JSLE is based on the American College of Rheumatology classification (revised criteria 1997).1

SLE has known effects on many organ systems, including musculoskeletal, renal, cutaneous, and cardiovascular systems. Patients with SLE have an increased risk of osteoporosis and fractures.2-4 The etiology of bone loss is multifactorial and can be attributed to systemic inflammation, metabolic and hormonal factors, medication-induced adverse effects, and clinical osteoporosis risk factors.2 Similar to the general population, factors such as increased age, postmenopausal status, smoking or alcohol use, low body weight or low body mass index are independent risk factors for osteoporosis in SLE.2,4 Patients with SLE can have additional risk factors for low bone mineral density (BMD) such as impaired renal function, longer disease duration, organ damage, markers of inflammation, low 25-hydroxyvitamin D serum levels, and increased number of childbirths.4

Glucocorticoids (GCs) are commonly used to treat SLE. The effects of GCs on bone mass are unclear.2,4 When used chronically, GCs induce bone loss; however, they also suppress systemic inflammation, which may have a beneficial effect on bone mass. Patients with SLE have a 1.2-4.7-fold increased risk of symptomatic fracture versus controls that have been age-matched and sex-matched. Risk factors for fracture include previous GC use, disease duration, and severity.2

Common variable immunodeficiency (CVID) is a disorder that impairs the immune system and is among the most common primary immunodeficiencies (affects 1/25 000). People with CVID have a deficiency of the antibodies IgG, IgA, and IgM.
A shortage of these antibodies makes it difficult to fight off infections, and these patients present with certain dental manifestations. Oral manifestations include a higher presentation of herpes sores, candidiasis, tonsillitis, gingivitis, calculus, enamel hypoplasia, and other ulcerations. Moreover, there is a greater risk for developing caries. Studies show more caries and higher gingival inflammation in patients with immunodeficiency than in control individuals. The increased incidence of dental caries can also be related to dental neglect, as the patients and their parents are focusing on the main disorder.5,6

Rehabilitation of severely resorbed maxillary and mandibular bone requires the aid of grafting procedures. Reports in the literature have shown that autologous onlay cortico-cancellous hip grafts in extreme maxillary resorption have a high cumulative success rate (95%) over a 10-year observation period.7 Conversely, reports on reconstruction of severely resorbed alveolar ridge using bovine bone xenograft for augmentation suggest a survival rate of 80.5%.8 Autogenous grafts provide osteogenic, osteoconductive, and osteoinductive properties.9 These grafts require primary watertight closure and vascularized soft tissue over the donor site.9 Typically, 10 cm³ of uncompressed bone is needed for every 1 cm of bony defect.9 With iliac crest grafts, morbidity includes lateral thigh paresthesia and immediate postoperative gait disturbance.9 Nerves at risk include the subcostal, iliohypogastric, and the lateral femoral cutaneous nerve.7

Temporary or transitional mini implants can stabilize a prosthetically driven therapy for SLE and CVID patient. These implants are useful for immediate fixation of the prostheses, and restoration for a complete oral rehabilitation. She reported a history of long-term glucocorticoid use as well as immunomodulators, leading to severe atrophy of the maxilla and mandible with malformed teeth and a very high caries incidence. Her lupus has been well controlled, and she has not been on glucocorticoids for several years. She receives intravenous immunoglobulin every 4 weeks for CVID.

On exam, she presented with multiple missing teeth, a severely atrophic maxilla and mandible, and remaining teeth with advanced carious lesions (Figure 1). A cone beam computed tomography (CBCT) scan showed bone loss, malformed teeth, impacted teeth, hypoplasia of the maxilla and mandible in the buccal-palatal dimension, and bilateral pneumatized maxillary sinuses (Figure 2). Radiographic and clinical exam revealed the remaining teeth to be non-restorable due to gross caries and loss of tooth structure. Treatment was planned with the Postgraduate Prosthodontic Department at Columbia University College of Dental Medicine for a full mouth rehabilitation.

Initial Surgical Stage

Under local anesthesia, her remaining erupted teeth were extracted, and immediate maxillary and mandibular complete dentures were delivered. An anterior iliac hip graft for the maxilla and mandible and bilateral maxillary sinus lifts with BMP-2 Infuse autologous bone graft was scheduled in the operating room. The impacted maxillary canines were extracted. The anterior iliac hip graft was harvested. A surgical stent was made by duplicating the patient's existing immediate dentures with clear auto curing methyl methacrylate resin (Caulk Orthodontic Resin, Dentsply, York, Pa). A surgical marker was used to mark the ideal location for the future implants on the ridge, which provided a guide as to where the bone from the hip graft should be positioned (Figure 3). The cortical bone from the hip graft was shaped to augment the buccal portion of the maxilla from first molar to first molar and secured using stabilization titanium screws (1.2-mm diameter, Stryker, Kalamazoo, Mich; Figure 4). Cancellous bone from the hip was used with BMP-2 Infuse (Medtronic, Minneapolis, Minn) to augment the maxillary sinus lifts. Cancellous autologous bone was also used as a particulate graft over the buccal aspect of the extraction site of the impacted right mandibular canine. Four maxillary and four mandibular transitional implants (1.8×7-mm Atlas implants, Dentatus, New York, NY) were placed in areas adjacent to future implant sites (Figure 5). In the operating room, the immediate dentures were relined with a resilient silicone reline material (Dentatus Tuf-Link, Dentatus) to fit the transitional implant ball attachments. Before suturing the soft tissue, the flanges of the dentures were eliminated to avoid transmucosal loading of the grafted areas (Figure 6). The patient was discharged on postoperative day one with pain medication, antibiotics, and a 0.12% chlorhexidine gluconate rinse. Postoperative panoramic radiograph shows the bone graft and the 8 transitional implants (Figure 7). The patient was seen for multiple postoperative visits to evaluate healing, adjust occlusal and intaglio surfaces of the prostheses, and reinforce oral hygiene. The postoperative course was uncomplicated (Figure 8).

CASE REPORT

Patient Selection

A 28-year-old woman with a history of juvenile-onset systemic lupus erythematosus and common variable immunodeficiency presented to the Division of Oral and Maxillofacial Surgery at Columbia University College of Dental Medicine (New York, NY) for an evaluation for dental rehabilitation. She reported a history of long-term glucocorticoid use as well as immunomodulators, leading to severe atrophy of the maxilla and mandible with malformed teeth and a very high caries incidence. Her lupus has been well controlled, and she has not been on glucocorticoids for several years. She receives intravenous immunoglobulin every 4 weeks for CVID.

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Implant Placement Post-Grafting

Four months later, the patient’s existing dentures were duplicated with clear self-curing methyl methacrylate resin (Caulk Orthodontic Resin, Dentsply) to fabricate a radiographic guide. The radiographic guide was stabilized on the transitional implants with a resilient silicone (Tuf-Link, Dentatus). The CBCT scan showed adequate augmentation of the bilateral maxillary sinuses as well as augmentation of the maxilla in the buccal-palatal dimension. The radiographic stents were then converted into surgical guides for the placement of 8 maxillary and 7 mandibular implants (Figure 9). Surgery was performed with intravenous sedation, and the patient received preoperative antibiotics intravenously along with dexamethasone to decrease postoperative swelling. All implants (Certain Tapered, Biomet 3i, Palm Beach Gardens, Fla) were placed with at least 35 N/cm insertion torque (Figure 10). The following fixtures were used: #2, 5×11.5 mm; #3, 4.1×10 mm; #4, 4.1×10 mm; #7, 4.1×8.5 mm; #10, 3.25×8.5 mm; #11, 3.25×8.5 mm; #13, 4.1×10 mm; #14, 4.1×11.5 mm; #19, 4.1×8.5 mm; #20, 3.25×8.5 mm; #21, 4.1×10 mm; #25, 4.1×11.5 mm; #28, 3.25×11.5 mm; and #30, 4.1×8.5 mm. The transitional implants were stable and maintained to support the transitional dentures to avoid transmucosal pressure to the newly placed implants. The postoperative panoramic radiographs showed the implants parallel to one another and with adequate anterior-posterior spread (Figure 11). Her postoperative course was uncomplicated.

Second Stage Implant Surgery

The implants were uncovered 3 months later. All implants were stable except the implant in the mandibular right first premolar, which was mobile and was removed and the area grafted. It was decided at this time to proceed to restore with the 6 remaining mandibular fixtures. The dentures were relieved in the areas contacting the healing abutments to avoid uncontrolled lateral transmucosal loading.

Prosthetic Rehabilitation

One month post second stage surgery, the patient presented for implant level impressions. Maxillary and mandibular open-tray fixture-level impressions were made using polyether impression material (Impregum Penta, 3M ESPE). Prior to the impression, all impression copings were splinted intraorally with self-curing methyl methacrylate resin (Pattern Resin LS, GC America, Alsip, Ill). Master casts were poured using Type IV dental stone (GC FujiRock EP, GC Europe, Kortrijk, Belgium) and verified with a verification jig made of self-curing methyl methacrylate resin (Pattern Resin LS, GC America). Record bases were fabricated using diurethane dimethacrylate resin (Custom Tray Material, Henry Schein, Melville, NY) and wax (TruWax, Dentsply) to obtain accurate interocclusal records and deter-
mine ideal lip support and occlusal vertical dimension of occlusion. The master casts were articulated using a facebow transfer on a semi-adjustable articulator (Artex CPR, Amann Girrbach, Koblach, Austria).

The initial diagnostic casts were scanned using digital technology along with both master casts and occlusal rims. Using Prosthetic Design Center (PDC, Stoneglass Laboratories, Homebush, Australia), a digital setup was completed to position the teeth in the ideal position based on the occlusal rims and the patient’s articulated pre-op diagnostic casts, copying the shape and size of the patient’s original teeth. Implant direction was then superimposed with the ideal tooth position to determine the screw access hole position and the need for angled abutments (Figure 12). A titanium bar was digitally designed and milled (Grade 5 Ti 6Al-4V). A 3D print of the setup was created that vacuum forms over the milled titanium frame (VisiJet M3 Stoneplast, 3D Systems, Rock Hill, SC) and tried intraorally for esthetics, phonetics, vertical dimension, and occlusion. The tooth try-in was verified, and fixed provisional prostheses were fabricated using the same titanium frame, tooth-colored acrylic (Palavit 55 VS, Heraeus Kulzer, South Bend, Ind) and pink-colored composite resin (Signum Cre-active, Heraeus) to create the artificial gingiva.

The transitional implants were removed by reverse torque and 30-degree angle correction transmucosal abutments were delivered on the maxillary left lateral incisor and canine implants, along with the maxillary and mandibular fixed provisionals. Four months following insertion of the fixed provisionals, the patient had no pain or discomfort on function and was satisfied with the esthetics. Maxillary and mandibular master casts were made and verified using the same technique described for the fixed provisional fabrication. To maximize the framework’s accuracy of fit before final digital scanning, a fit test was performed using a plaster key. This key was prepared by joining non-engaging titanium temporary cylinders with impression plaster. The fit test was performed intraorally. In the absence of passivity, the fragility of the plaster would cause it to fracture.

The articulated master casts and provisional casts were scanned using the PDC System. The final titanium framework width, height, and thickness of preparation struts was designed to support each individual pressed lithium disilicate crowns (Prosthetic Design Centre software, Stoneglass; Figure 13). The crowns were fabricated from printed patterns digitally designed and individually cemented on the titanium framework (Figure 14) with opaque resin cement (Multilink Automix, Ivoclar Vivadent, Amherst, NY). Pink-colored composite resin

Figure 2. Pre-operative cone beam computer tomography scan.
FIGURES 3–5. FIGURE 3. (a) Surgical guide with surgical marker for ideal position of the implants. (b) Existing bone ridge with surgical mark of ideal implant position. FIGURE 4. (a) Sectioning of the cortical hip bone. (b) Stabilization of the bone graft with stabilization titanium screws. FIGURE 5. Maxillary occlusal view after placement of transitional implants.
(Signum Cre-active, Heraeus) was applied to create the artificial gingiva.

Final restorations were delivered after verification of fit clinically and radiographically (Figure 15). The prostheses were torqued to manufacturer’s recommendations and access holes sealed with polytetrafluoroethylene film (Teflon tape) and packable composite (Filtek, 3M ESPE; Figure 16). A maxillary occlusal guard (Drufosoft, Dentsply) was given to the patient to wear at nighttime. The patient was placed on 4-month recall and has been seen for over 18 months without any surgical or prosthetic complications.

**FIGURES 6–9.** Figure 6. (a) Intaglio view of maxillary interim complete denture. Note complete flange reduction and resilient silicone reline. (b) Frontal view in maximum intercuspation of maxillary and mandibular interim complete dentures adjusted and delivered after surgery. Figure 7. Postoperative panoramic radiograph. Figure 8. Intraoral frontal view of transitional implants 4 months after healing. Figure 9. Placement of maxillary implants using the stabilized surgical guide.

**DISCUSSION**

SLE is a disease with a wide range of signs and symptoms, including decreased bone mineral density. The etiology of bone loss is multifactorial and is due to systemic inflammation, metabolic factors, hormonal factors, medication-induced adverse effects, and clinical osteoporosis risk factors. The young woman patient presented with missing and malformed teeth, multiple carious lesions, and severe atrophy of the maxilla and mandible. Her medical history was also complicated by common variable immune deficiency, leading to bronchiectasis and frequent respiratory infections. Communications with her immunologist and pulmonologist was essential to planning her
dental reconstruction. The patient was highly motivated and aware of the length of time to complete her treatment, as well as the risks of bone graft failure, need for multiple surgeries, and grafting procedures. Her postoperative course was uncomplicated, and she had excellent success with the grafting procedures. The alternative treatment plan would include removable prostheses with or without implants; however, due to the atrophic maxillary and mandibular ridges, removable prostheses without implants would not have been an optimal alternative and would have led to further bone resorption. Due to the extent of bone augmentation, the alternative of an immediate load case on implants was contraindicated. By having a comprehensive treatment plan and sequence with emphasis on a well-executed interdisciplinary approach, the case was staged with minimal complications, ensuring case control throughout the course of the treatment.

The autogenous iliac bone graft used allowed for a sufficient amount of donor bone to reconstruct both the severely resorbed maxilla and mandible with one donor site. Extraoral bone harvesting was considered superior to intraoral bone harvesting due to the deficiency of bone in the symphysis and ramus areas, as well as the volume of bone needed for reconstruction. Cortico-cancellous blocks, as well as abundant cancellous bone, were easily harvested and used to reconstruct this patient’s defects. The low risk of morbidity associated with anterior iliac hip harvesting allowed this patient to quickly recover and return to her normal routine after surgery. The patient did not suffer from any postoperative neurological damage and was ambulatory the day after surgery.

The extraction of the patient’s remaining dentition and adjustment to complete dentures could have been an emotionally difficult time for the patient. The transitional implants aided in the retention and stability of the dentures, which improved speech and mastication. Moreover, the implants improved the psychological acceptance of the complete dentures during the healing time for the bone graft and implant osseointegration. Additionally, the transitional implants enabled the grafted areas to heal without occlusal loading.

The quality of life, functional, and esthetic outcomes were assessed relative to risks. In an extensive review of the literature, Diz et al found that control of the systemic disease was more important than the specific disease itself. In this case report, control of the disease was necessary prior to surgical intervention. Careful interdisciplinary planning allowed proper implant placement position and a straightforward rehabilitation with case control throughout the course of treatment. The use of the patient’s preoperative records to mimic the position, shape, and size of her natural teeth on the final restoration provided an esthetic and emotionally satisfying result. The treatment techniques and sequence used in this case can be applied to other complex rehabilitation cases.
**Figure 12.** Screenshot of digital planning after determination of implant and teeth position.

**Figure 13.** Screenshot of digital planning of titanium framework for final maxillary restoration.
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NOTE

The authors have no conflicts of interest.

REFERENCES