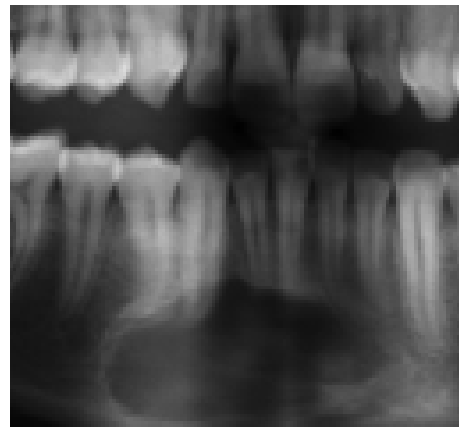
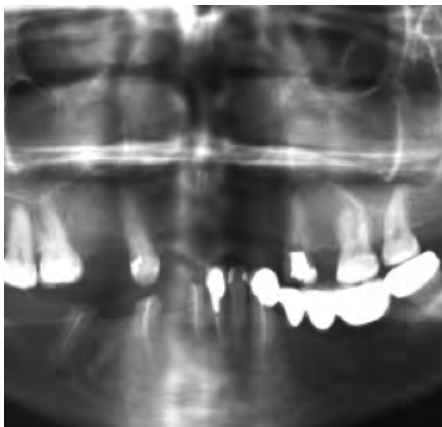
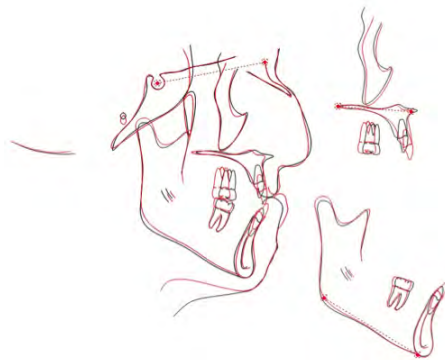
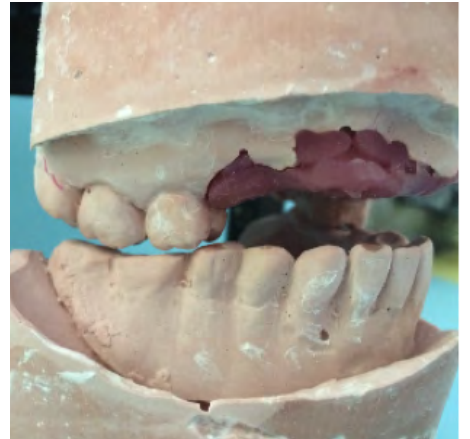
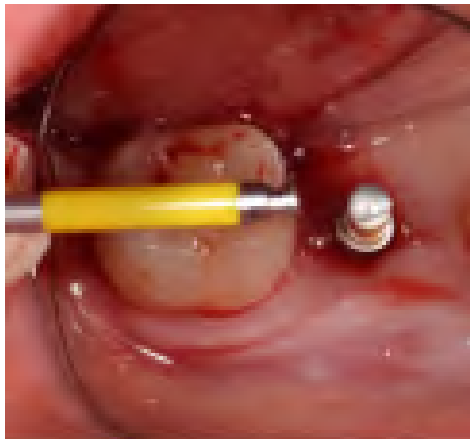


CDR

COLUMBIA DENTAL REVIEW



CDR



COLUMBIA UNIVERSITY
College of Dental Medicine

COLUMBIA DENTAL REVIEW 2015-2016

Editorial Board

Editor-in-Chief

Alina O'Brien '17

Assistant Editors

Student Liaisons

Mary Awadallah '17

Jonathan Lomboy '17

Draft Editors

Ryan Donelan '17

Cathy Lee '18

John Lankalis '17

Final Editors

Michael Fogge '17

Kevin Lee '17

Design and Layout

Andrea Kim '17

Jessie Yang '17

Alan Kuo '17

Keyur Naik '17

Faculty Advisor

Letty Moss- Salentijn, DDS, PhD

Faculty Reviewers

Thomas Boyle, DMD, ABGD

Vicky Evangelidis, DDS, MPH

June Harewood, DDS

Shantanu Lal, DDS

Louis Mandel, DDS

Letty Moss-Salentijn, DDS, PhD

Elizabeth Philipone, DMD

Angela Yoon, DDS, MPH, MAMSc

Aim and Scope

The Columbia Dental Review (CDR) is an annual publication of Columbia University College of Dental Medicine (CDM). This journal is intended to be a clinical publication, featuring case presentations supported by substantial reviews of the relevant literature. It is a peer-reviewed journal, edited by the students of the school. The editors are selected on the basis of demonstrated clinical scholarship.

Authors are primarily CDM students from pre-doctoral and post-doctoral programs, CDM faculty and residents, and attendings from affiliated hospitals. Peer reviewers are selected primarily from the CDM faculty. Submissions undergo a blind peer review system whereby the authors are not known by the reviewers (at least two per manuscript). Instructions for authors wishing to submit articles for future editions of the CDR can be found on the last page of this journal. Opinions expressed by the authors do not necessarily represent the policies of Columbia University College of Dental Medicine.

Editors' Note

Dear Readers,

I am delighted to welcome you to the 2015-2016 edition of the Columbia Dental Review. The College of Dental Medicine has a long history of producing excellent research, and the goal of the Review is to share some of the innovative and collaborative work that take place at our school. Thank you to our team editors for their hard work, and I hope you enjoy the issue.

Sincerely,

Alina O'Brien '17

Table of Contents

Dental implant surgery and restoration on a patient with von Willebrand disease: A case study Michael Kang, DDS, Phillip Y. Kang, DDS	1
Partial Mouth Reconstruction with Combined Fixed and Removable Prosthesis: A Case Report Jessica Wyatt, DDS, Francis Oh, DDS, MS, MA, PhM	4
Severe Periodontal Abnormalities in Systemic Sclerosis (Scleroderma): A Case Report Jaffer A Shariff, DDS, MPH, Philip Y. Kang, DDS	8
Traumatic Bone Cyst in a Child: A Case Report Gloria Lee, DDS, Katrin Tamari, DDS	11
Treatment of an Adult Unilateral Complete Cleft Lip and Palate Patient: A Case Report Keith Jaffe Goldman, DMD, MS, Jing Chen, DDS, PhD	14

Dental implant surgery and restoration on a patient with von Willebrand disease: A case study

Michael Kang, DDS¹, Phillip Y. Kang, DDS²

¹Periodontic Resident, College of Dental Medicine, Columbia University, New York, NY

²Assistant Professor of Dental Medicine, Division of Periodontics, College of Dental Medicine, Columbia University, New York, NY

Abstract

Von Willebrand disease is not commonly encountered in practice, but is actually the most common hereditary disorder affecting coagulation. A mutation inhibiting von Willebrand factor, which is a key clotting protein that binds to factor VIII, affects platelet adhesion during wound healing. Patients with this disorder are at a higher risk of postoperative complications following dental surgery, but can still be treated successfully. Currently, desmopressin or DDAVP is the method of treatment for patients with Willebrand disease in preparation for surgery. DDAVP stimulates the release of von Willebrand factor from endothelial cells, and can be administered intranasally or intravenously. The following case study documents a young 21-year old patient with Von Willebrand disease that had a dental implant placed to restore a missing tooth. Through collaboration with a hematologist administering prophylactic DDAVP, the implant surgery was performed without any postoperative complications. The implant successfully integrated and was restored into function with no unusual clinical manifestations. The case study also details additional precautions taken during treatment planning to minimize the patient's need for surgical procedures during therapy by avoiding a two-stage protocol and by utilizing a tissue level implant design.

Introduction

Von Willebrand disease (vWD) is the most common hereditary bleeding disorder.¹ A mutation inhibiting von Willebrand factor, which is a key clotting protein that binds to factor VIII, affects platelet adhesion during wound healing.² Patients affected by von Willebrand disease present with degrees of excessive bleeding, which may manifest in frequent nosebleeds, bleeding gums, and bruising. Female patients affected can have heavier menstrual periods. An uncommon symptom is severe internal bleeding or hemarthrosis.¹ Clearly, vWD can pose a significant problem in a patient indicated for surgical procedures intraorally. Cases of dental surgery and extractions on vWD patients have been documented in the past, and complications with excessive gingival bleeding pose a concern.^{3, 4, 5, 6}

Managing excessive bleeding is nothing new in the field of medicine, and in particular, several methods have been shown to be effective for dental surgery.³ Local application of tranexamic acid (TA) and fibrin glue have been used in past cases to provide hemostasis prior to the procedure. TA acid is often used in major trauma cases at hospitals, as well as on hemophiliacs with excessive bleeding during dental procedures.⁴ It is typically delivered as a mouthwash for dental applications, but can be given orally or intravenously.

TA acid is a synthetic derivative of lysine, and it binds to the lysine receptors on plasmin molecules to prevent the degradation of plasmin to fibrin. Fibrin glue is also a commonly used to manage bleeding complications.⁵ Fibrin glue is composed of fibrinogen and thrombin, derived from human and bovine sources, and is used to help create a fibrin clot.

Specific for von Willebrand disease, desmopressin (DDAVP) has been used with success in managing bleeding after surgery.⁶ Desmopressin has a number of different effects upon administration. The most applicable to patients with vWD is that DDAVP stimulates the release of von Willebrand factor from endothelial cells, which can promote the clotting cascade from occurring. Typically, DDAVP is administered intravenously at a dosage of 0.3 ug/kg in a 50 mL 0.9% saline solution. The DDAVP should be administered at least one hour prior to any surgical procedure, and lasts for about 8 to 10 hours.⁷

Case Report

A 21-year old female patient with von Willebrand disease presented in the Columbia University College of Dental Medicine clinics for comprehensive care. She had lost #19 due to caries, which was previously extracted about one year prior (Figure 1). The optimal treatment plan was determined to be a single tooth implant.

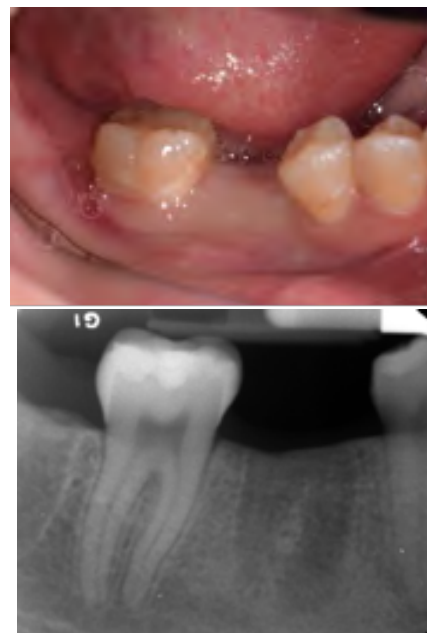


Figure 1 Preoperative clinical photograph and periapical radiograph of #19

Dental implant surgery and restoration on a patient with von Willebrand disease: A case study

Previously, #19 was extracted after the patient received a single dose of desmopressin (DDAVP) from her hematologist, and the procedure was successful with no adverse events or complications with bleeding. After consulting with the patient's hematologist, it was again recommended that DDAVP be administered approximately one hour prior to the start of implant surgery. An hour prior to implant surgery, the patient received approximately 0.3 ug/kg of DDAVP in a 50 mL 0.9% saline solution intravenously.

Additional accommodations were made to minimize the amount of surgeries and traumatic events to the tissue that would induce bleeding. A tissue level implant was used to eliminate second stage surgery and to provide adequate biological width away from the healing abutment to prevent bleeding during prosthetic visits. A flapless surgical technique was also used to prevent a full thickness elevation of the surrounding soft tissue during the procedure (Figure 2). The ridge was assessed clinically and with a computerized tomography radiograph to ensure adequate width of bone with minimal concavities.



Figure 2 Implant surgery, flapless approach using Straumann 4.8 x 10 mm tissue level implant

No postoperative complications were noted, and the patient reported no issues of excessive bleeding following the surgery. The implant was allowed four months for healing, and was restored (Figure 3).



Figure 3 Clinical photograph and periapical radiograph of #19 final restoration, completed four months after implant placement

Discussion

Successful treatment of patients with von Willebrand disease, and other hemophiliac disorders, requires collaboration with the patient's hematologist for the appropriate method of treatment. Many successful cases of surgical procedures on patients with hematological disorders can be treated safely with minimal complications with proper intervention and treatment planning.⁸ In this particular case, the risk was minimized through careful collaboration with a hematologist and prophylactic DDAVP, but measures were taken in the surgical approach and the implant design as well.

Flapless approach to implant surgery has been shown to be successful, provided that the appropriate cases are selected.⁹ If adequate horizontal bone is present with minimal concavities present apically, a flapless approach provides a conservative approach with minimal risk of perforations. A cone beam computerized tomography scan and the use of a surgical guide is recommended if no flap is reflected to allow for visualization of the bone as the implant

Dental implant surgery and restoration on a patient with von Willebrand disease: A case study

is placed. In the case documented, the flapless approach allowed for a minimally invasive approach and limited postoperative bleeding from the surgery.

Although bleeding soft tissue trauma can be managed successfully, the use of dental implant design can also minimize trauma in both the surgical and prosthetic visits.¹⁰ Typically, the process of receiving a dental implant can involve multiple procedures that induce bleeding in the soft tissue. The most traumatic procedure involves the actual placement of a dental implant or the second stage surgery to attach a healing abutment after integration. However, bone level implant designs are more susceptible to incidents of soft tissue trauma during the prosthetic phase. Irritation from prosthetic handling of the healing abutment, or dislodgement of the healing abutment, can result in soft tissue trauma and a necessity to redo second stage surgery for bone level implants. In one-stage surgical approaches, epithelium can adhere to healing abutments and result in trauma and bleeding on removal after implant integration.¹¹ For these factors, a tissue level implant was used to limit incidents of bleeding during prosthetic fabrication.

This case study follows one example of treatment involving a patient with von Willebrand disease, but the techniques and approaches cannot be extrapolated to every case. The patient history and clinical parameters must be taken into account for each individual case. Additional clinical trials are needed to determine the optimal approach for patients with hematological disorders. However, on the basis of existing literature, many cases with patients that have von Willebrand disease and other hematological deficiencies can be treated successfully with limited complications.

Conclusion

A patient with von Willebrand disease was successfully treated for implant surgery and restoration with no complications. Communication with the patient's hematologist was a key component of successful therapy. Several methods of hemostasis control and careful selection of implant type can aid in providing a successful result and minimize the amount of trauma.

References

1. Lillicrap, D. (2013). von Willebrand Disease: Advances in Pathogenetic Understanding, Diagnosis, and Therapy. *American Society of Hematology*, 122: 3735-3740.
2. Sadler JE, B. U. (2006). Update on the pathophysiology and classification of von Willebrand disease: a report of the Subcommittee on von Willebrand Factor. *Journal of Thrombosis and Haemostasis*, 4: 2103-2114.
3. van Galen KPM, E. E.-B. (2015). Antifibrinolytic therapy for preventing oral bleeding in patients with haemophilia or

Von Willebrand disease undergoing minor oral surgery or dental extractions (Review). *Cochrane Database of Systemic Reviews*.

4. Dunn CJ, G. K. (1999). Tranexamic acid: a review of its use in surgery and other indications. *Drugs*, 57(6): 1005-10032.
5. Whitman DH, B. R. (1997). Platelet gel: An autologous alternative to fibrin glue with oral applications in oral and maxillofacial surgery. *Journal of Oral and Maxillofacial Surgery*, 1294-1299.
6. Federici AB, S. R. (2000). Optimizing local therapy during oral surgery in patients with von Willebrand disease: effective results from a retrospective analysis of 63 cases. *Hemophilia*, 6: 71-77.
7. Sanchez-Luceros A, M. S. (2010). Biological and clinical response to desmopressin (DDAVP) in a retrospective cohort study of children with low von Willebrand factor levels and bleeding history. *Blood Coagulation, Fibrinolysis, and Cellular Haemostasis*, 984-989.
8. Zulfikar B, K. B. (2016). Surgery in patients with von Willebrand disease. *Blood Coagulation and Fibrinolysis*, 1-5.
9. Campelo LD, C. J. (2002). Flapless implant surgery: A 10-year clinical retrospective analysis. *International Journal of Oral and Maxillofacial Implants*, 17: 271-276.
10. Nickles K, W. M. (2010). Comprehensive Treatment of Periodontitis in Patients with von Willebrand Disease. *Journal of Periodontics*, 81: 1432-1440.
11. Hermann JS, C. D. (2001). Biological Width around one- and two-piece titanium implants. *Clinical Oral Implants Research*, 6: 559-571.

Partial Mouth Reconstruction with Combined Fixed and Removable Prostheses: A Case Report

Jessica Wyatt, DDS¹, Francis Oh, DDS, MS, MA, PhM²

¹Class of 2016, College of Dental Medicine, Columbia University, New York, NY

²Assistant Professor of Dental Medicine, Division of Prosthodontics, College of Dental Medicine, Columbia University, New York, NY

Abstract

Partial and full mouth reconstruction can present multiple challenges to the dentist. However, the benefits that result from this reconstruction can be life-changing for the patient. This case presents a 53-year-old female who presented with the chief complaint that she had “a broken bridge” and wanted to “replace all of her missing teeth.” Treatment for this patient consisted of combined fixed and removable prostheses: a 10-unit implant-supported maxillary fixed partial denture (FPD) and an implant-supported complete mandibular overdenture.

Case Report

A 53-year-old female presented to Columbia University Dental Clinic with the chief complaint that she had a “broken bridge” and would like “to replace all of her missing teeth.” Periapical and panoramic radiographs were taken on this patient (Figures 1a-b) and she proceeded to the senior clinic for a comprehensive exam, photos (Figure 2), and treatment planning session.



Figure 2 Initial extra- and intra-oral photographs

Clinical exam revealed multiple missing maxillary anterior teeth and recurrent decay of remaining anterior maxillary teeth. The patient’s chief complaint related to the FPD on the lower right quadrant (25-X-27-X-29) fractured in the area of tooth #29. Periodontally, pockets were within normal limits but oral hygiene appeared to be very poor. Radiographs revealed a periapical radiolucency at the apex of tooth #12 and multiple root fragments remained in the mandibular anterior region. After discussion with the patient, she expressed that she currently used removable prostheses but was interested in restoring her edentulous areas with fixed prostheses, such as implants.

A CBCT scan was taken on the patient and the finalized treatment plan was created consisting of a maxillary implant-supported partial overdenture and mandibular implant-supported FPD to restore occlusion in the lower right quadrant where the patient’s previous FPD was located. The patient’s main concern related to finances, which limited her to restoring only half of the mandibular arch at this time, and later, restoring the other half.

Over a period of 4 months, the patient had extractions of teeth #6, 12, 17, 22, 23, 24, 25, 27, 29, Straumann implants placed in sites #4, 6, 9, 13, 22, 27, and Maxxeus bone grafts placed in sites #6, and 22.

It was decided, together with the senior dentist and faculty, to update the treatment plan to: maxillary 10-unit implant-supported FPD with complete mandibular implant-supported overdenture. An interim mandibular complete denture was fabricated for the patient to wear while the mandibular bone graft continued to heal.

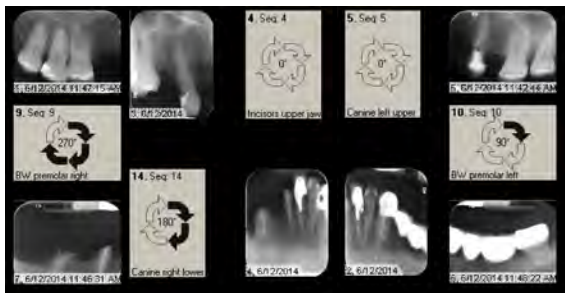


Figure 1a Initial periapical radiographs



Figure 1b Initial panoramic radiograph

Partial Mouth Reconstruction with Combined Fixed and Removable Prosthesis: A Case Report

The patient was provisionalized initially with a lab-fabricated acrylic FPD that rested on teeth #6 and 12, before they were extracted, to allow for the bone graft placed near site #6 and 27 to heal. After teeth #6 and 12 were extracted, the patient was given an interim maxillary partial denture and an interim mandibular complete denture, both with relief in the areas of the implant healing abutments.

The next step, to help establish proper lip support and vertical dimension of occlusion (VDO) in fabricating the final prosthesis, was lab-fabricated provisional for the maxillary implant supported FPD. An addendum to the existing treatment plan was made for this provisional.

Approximately one year after maxillary implant placement, the patient presented for the final restoration. Healing abutments were removed and impression copings were placed in the maxillary implant sites. A GC resin pattern jig, supported by floss, was created to capture each implants angulation relative to the others and a maxillary vinyl polysiloxane (VPS) open-tray impression was taken over the implants and jig (Figure 3a-b). A soft tissue model was poured up and the GC resin pattern jig with the impression copings was seated on the cast and was reinforced with more GC resin pattern.

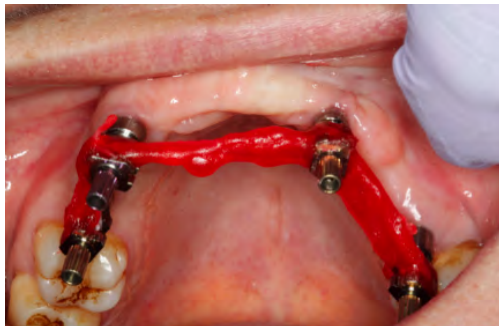


Figure 3a GC resin pattern/floss jig



Figure 3b Open-tray VPS impression over jig

The jig was then placed in the patient's mouth (Figure 4a-b) to verify that the cast was an accurate reproduction of the implant angulation present in the patient's mouth.



Figure 4a Reinforced jig seated in cast



Figure 4b Reinforced jig in patient's mouth

Mandibular pick-up impression of the patient's interim mandibular complete denture was taken in alginate and a cast was poured up. Putty bite registration was taken and the maxillary and mandibular casts were mounted on an articulator (Figure 5) The mounted casts were sent to the lab for fabrication of the maxillary custom abutments, provisional FPD, and metal framework for the final FPD. A slight open bite was present in the posterior right side, as the interim mandibular denture was loosely retentive and thus, was not entirely accurate in mounting a perfect bite (Figure 6). However, this was acceptable since a new complete mandibular denture was to be fabricated against the final completed maxillary FPD. Shade B1 was selected together with the patient for the lab-fabricated maxillary provisional.

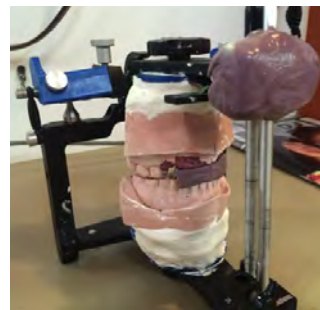


Figure 5 Mounted casts with putty bite

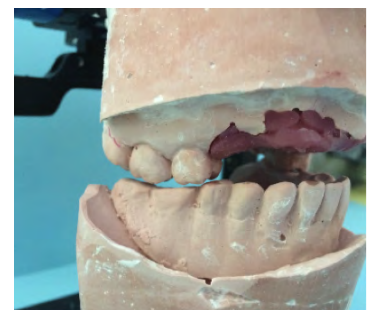


Figure 6 Mounted casts, posterior open bite

The patient returned and the 4 maxillary custom abutments were torqued down. The lab-processed maxillary acrylic provisional was tried in, adjusted, and relined over a series of three appointments, allowing the patient to adjust to the provisional and provide feedback on esthetics and functional concerns. The patient also expressed that she wanted "all of

Partial Mouth Reconstruction with Combined Fixed and Removable Prosthesis: A Case Report

the teeth to be the same length" incisally, so incisal adjustments were made as such. The patient, despite having a relatively low smile line, was concerned with the cervical overextension of the teeth, particularly tooth #10. After discussion with the patient and faculty, the decision to eventually add a continuous band of pink porcelain to the final prosthesis was made.

During the provisional-adjustment appointments, the maxillary metal framework for the final prosthesis was tried in and adjusted. Initially, the metal framework appeared to have a cant (Figure 7a). Using the patient's interpupillary line as a paralleling reference point, the metal framework was adjusted (Figure 7b).



Figure 7a Metal framework demonstrating cant



Figure 7b Framework after adjustment

Upon further examination, it appeared that the maxillary provisional's midline was canted. Aside from this, the patient was satisfied with the esthetics and functionality of the acrylic maxillary provisional and a VPS pick up impression of the adjusted maxillary metal framework and an alginate pick up impression of the acrylic provisional were taken. A new putty bite registration with a wooden stick was taken against the maxillary provisional and patient's interim complete denture. The wooden stick was adjusted so that it was parallel to the patient's interpupillary line. The alginate impression was poured up and the cast, the new bite registration, and the previously mounted mandibular cast were sent to the lab for fabrication of the final maxillary prosthesis. Careful instructions were made to the lab to fabricate the final prosthesis with the new, correct, bite registration and to only use the cast of the maxillary provisional as an esthetic guide for the size and shape of the final prosthesis, since it was canted. The lab was also instructed to add a continuous band of pink porcelain to simulate gingiva, as discussed earlier.

The lab returned the bisque-baked maxillary final FPD,

which was tried in and adjusted using shim stock and Fit Checker ®. Pink porcelain was absent in the area of tooth #13 and very minimal in the area of tooth #4 (Figure 8). The patient was dissatisfied with this, and the final prosthesis was returned to the lab for addition of more pink porcelain in this area and for final glaze. No other esthetic concerns were raised by the patient at this time.



Figure 8 Bisque-baked final maxillary FPD demonstrating lack of pink porcelain tooth #13

The lab returned the final glazed maxillary FPD, which was tried in and minor adjustments were made. The patient was still dissatisfied with the lack of pink porcelain in areas #4 and 13, and with the amount of metal showing from the abutments, despite her low smile line. The patient was informed that more pink porcelain could not be added to the final prosthesis as overcooking in the oven might warp the underlying metal framework. The patient was then informed that other means to cover the exposed metal could be taken at a later time, such as adding gingival composite to these areas, etc. In the end, these are likely compromises to the true desired esthetic result which should have been addressed earlier on.

The screw holes in the custom abutments were protected with Fermit and the final maxillary restoration was cemented with conservative amounts of TempBond. Unfortunately, the patient reported later that evening that the final prosthesis had fallen out.

The patient returned the following day, minor adjustments were made to areas of pink porcelain that were overextended, and the final prosthesis was cemented with Improv implant cement.

As the maxillary prosthesis was being attended to, the final mandibular complete denture was also being worked on. The final complete denture was delivered, occlusion was adjusted, and it was converted to an implant-supported overdenture (Figure 9). The patient was satisfied with esthetics and function of both the maxillary and mandibular prostheses.

Partial Mouth Reconstruction with Combined Fixed and Removable Prosthesis: A Case Report



Figure 9 Pre- and post-treatment intra-oral photographs

Discussion

The use of a GC resin-pattern jig used during impressions has been shown in several studies to improve the accuracy of the master cast that is poured (1) but in other similar studies, the use of a resin-based jig, compared to other materials, such as plaster, showed no advantage in regards to final cast accuracy.

Though the patient was mostly edentulous upon initial presentation to the clinic and several of her teeth had poor prognosis, a few alternative treatment plans were presented; first, the option of both maxillary and mandibular removable dentures were presented to the patient. However, the patient, having already had experience with a very non-retentive mandibular complete denture, due to a lack of alveolar crest height, declined this option in favor of fixed prostheses.

Another option for restoring the mandibular dentition with implants that was presented to the patient was the option of the Nobel Biocare “All-on-Four” prosthesis (Figure 9). Unfortunately, due to financial constraints, the patient was unable to move forward with this option at the time. Initially, the patient did express interest in restoring the lower right quadrant with an implant-supported FPD, and later, restoring the lower left quadrant similarly, but this too became a financial issue that led the patient to select the option of the implant-supported mandibular overdenture instead. Still, the patient expressed that she would like to eventually replace the overdenture with a fixed prosthesis in the future.

Conclusion

Full and partial mouth reconstruction can be a challenging endeavor for both the dentist and the patient. Certain aspects of treatment may limit treatment options such as finances, bone quality, patient expectations, but with detailed planning and discussions with the patient, dramatic and successful results can be achieved.

References

1. Lee, Sang-Jik, and Sung-Bum Cho. “Accuracy of Five Implant Impression Technique: Effect of Splinting Materials and Methods.” *The Journal of Advanced Prosthodontics* 3.4 (2011): 177–185. *PMC*. Web. 19 Apr. 2016.

2. Hariharan, R., C. Shankar, M. Rajan, MR Baig, and NS Azhagarasan. “Evaluation of Accuracy of Multiple Dental Implant Impressions Using Various Splinting Materials.” *Int J Oral Maxillofac Implants*. (Jan-Feb 2010): 38-44. *PubMed*. Web. 02 Feb. 2016.

Severe Periodontal Abnormalities in Systemic Sclerosis (Scleroderma): A Case Report

Jaffer A Shariff, DDS, MPH¹, Philip Y. Kang, DDS²

¹Periodontic Resident, College of Dental Medicine, Columbia University, New York, NY

²Assistant Professor of Dental Medicine, Division of Periodontics, College of Dental Medicine, Columbia University, New York, NY

Abstract

Systemic sclerosis (Scleroderma) is an autoimmune or connective tissue disorder that is characterized by fibrosis of skin, muscles, blood vessels, and other organs such as lungs, kidney and gastrointestinal tract. Scleroderma mainly affects women between the ages of 30 to 50 years and has shown to affect facial and oral structures, such as salivary glands, taste buds, jaw, teeth and periodontium. It can cause limited mouth opening (microstomia), dry mouth (xerostomia), malocclusion, mouth sores, mouth stiffness, jaw pain, periodontal diseases, and tooth loss. This paper reports on the effect of scleroderma on the periodontium of a 39-year-old female.

Introduction

Systemic sclerosis (SSc), or Scleroderma, is an autoimmune or chronic connective tissue disease. It is characterized by excessive production and deposition of collagen within the skin and internal organs and is associated with vascular abnormalities and autoantibody production ⁰ in.^{1,2} The etiopathogenesis of SSc is unclear; potentially contributing factors include the presence of autoimmunity, genetic predisposition, certain environmental exposures and chance (possibly random genetic mutations).³ SSc is divided into two subsets of disease, limited or diffuse. The limited form is characterized by thickening of the skin confined to the areas distal to the elbows and knees, and is associated with less severe internal organ involvement. The diffuse form involves skin thickening proximal to the elbows and knees in addition to distal area involvement and is associated with more severe organ damage.² Epidemiological studies conducted in the United States since the 1989 have shown an annual incidence rate for scleroderma of 13.9 to 21 per million,^{4,5} with female to male ratio of 1:3.⁶

The orofacial region is commonly involved in SSc. Oral manifestations include skin and oral mucosal atrophy, limited mouth opening, mandibular resorption, and periodontal ligament space (PLS) widening.^{7,8} This paper reports a case of severe periodontal impairment associated with systemic sclerosis.

Case Report

A thirty-nine-year-old female presented with a chief complaint that her upper front teeth were loose, and the gums were sore and bleed on touch. Patient stated that she had SSc since birth and is currently under the care of a physician. She is being treated for SSc related skin and eye conditions. The patient also reported that she has had a

routine (every 6 months) scaling and root planning appointments for the past 3 years without any surgical or long-term treatment interventions. No other relevant medical conditions were reported. No history of drug or latex allergies were reported, with the exception of seasonal allergies due to pollen.

Her medications included topical steroids for the management of SSc related skin conditions and occasional use of analgesics (ibuprofen) and anti-histamines (Clarinet - desloratadine) for allergies.

The extraoral examination revealed stiffened (fibrosed), smooth, shiny, stretched, pale facial skin with hard and tightened oval and straight patches. The lips were smooth, rigid, inflamed and ulcerated at the corner of the lip (angular cheilitis) with a prominent loss of vermilion border of upper lip thereby leading to reduced mouth-opening (microstomia) (Figure 1). Hyperpigmentation and blanching around the face, neck, hand and feet was also noticed.



Figure 1a Loss of vermilion border

Figure 1b Microstomia

Figure 1c Angular cheilitis

Intraorally, >80% of her posterior teeth (molars and premolars) were missing. Acrylic pontic and splinting was present to replace the missing lower left central incisor. Spacing or diastemas were present in the upper anterior arch. Yellowish to brown stains were present on all remaining teeth surfaces. The buccal mucosa was thick and stiff with white patches and ulcerations (Figure 2). There was considerable narrowing of the maxillary palatal arch with the presence of diffused, non-scrapable white patches (Figure 2).

Severe Periodontal Abnormalities in Systemic Sclerosis (Scleroderma): A Case Report

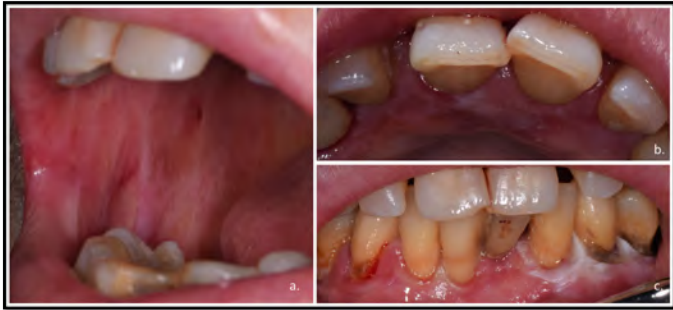


Figure 2a Buccal cheek stiffness with ulceration

Figure 2b, 2c White patches and diastemas

Periodontal changes:

The most astonishing finding was the absence of attached gingiva around all the upper teeth along with profuse bleeding on touch, as well as the absence of upper vestibule and shallowing of the lower vestibule (Figure 3). The alveolar mucosa of the upper arch appeared to have replaced the attached gingiva and was attached directly to the tooth surface (Figure 3). The full mouth periodontal evaluation showed generalized probing depths and a clinical soft tissue attachment loss ranging from 5mm to 12 mm, with severe bleeding on probing. Grades 1 to 2 mobility as per Millers classification⁹ were present in all upper teeth and grade 1 mobility was present in the remaining lower teeth. A few anterior teeth showed signs of occlusal trauma or instability (fremitus) caused by pathological tooth migration. Generalized plaque and calculus deposits were present. Similar diffused, non-scrapable, white patches observed on the buccal and palatal mucosa were also present on the alveolar mucosa and on the remaining band of attached gingiva around all upper and lower teeth (Figure 2).



Figure 3 Absence of keratinized/attached gingiva

Intraoral periapical radiograph and orthopantomogram revealed widening and thickening of periodontal ligament (PDL), with generalized vertical and horizontal bone loss in both upper and lower arches.

Discussion

Systemic sclerosis (SSc) is a generalized disorder of small arteries, micro-vessels and connective tissue, characterized by fibrosis and vascular obliteration in the skin, gastrointestinal tract, lungs, heart, and kidneys.¹⁰ In the

present case report, the extra oral, intraoral -soft and -hard tissue, and periodontal manifestation of SSc were similar to those described in other reports.^{11,12,13} However, while other case reports have mentioned a loss or reduction of attached gingiva and the vestibule, this report shows a complete loss or absence of attached gingiva and vestibule. The severity of the periodontal manifestation, along with microstomia, further limited the patient's ability to perform oral hygiene and lead to exacerbation of the already existing periodontal disease and soft tissue lesions.

A comprehensive treatment plan involving procedures such as, scaling root planing, soft tissue procedure (vestibuloplasty, connective tissue augmentation), and prosthetic reconstruction (with and without implants), is currently being considered.

References

1. Handin, R. I., Braunwald, E., Fauci, A., Kasper, D. L., Hauser, S., Longo, D. L., & Jameson, J. L. (2005). Harrison's principles of internal medicine. *Harrison's Principles of internal medicine*.
2. Barnes, J., & Mayes, M. D. (2012). Epidemiology of systemic sclerosis: incidence, prevalence, survival, risk factors, malignancy, and environmental triggers. *Current opinion in rheumatology*, 24(2), 165-170.
3. Russo, P. A., Lester, S., & Roberts-Thomson, P. J. (2014). Systemic sclerosis, birth order and parity. *International journal of rheumatic diseases*, 17(5), 557-561.
4. Steen, V. D., Oddis, C. V., Conte, C. G., Janoski, J., Casterline, G. Z., & Medsger, T. A. (1997). Incidence OF systemic sclerosis in allegheny county, Pennsylvania. A twenty-year study of hospital-diagnosed cases, 1963–1982. *Arthritis & Rheumatism*, 40(3), 441-445.
5. Mayes, M. D., Lacey, J. V., Beebe-Dimmer, J., Gillespie, B. W., Cooper, B., Laing, T. J., & Schottenfeld, D. (2003). Prevalence, incidence, survival, and disease characteristics of systemic sclerosis in a large US population. *Arthritis & Rheumatism*, 48(8), 2246-2255.
6. Gilliland, B. C. (1998). Systemic sclerosis (scleroderma). *Harrisons Principles of Internal Medicine*, 1888-1895.
7. Stafne, E. C., & Austin, L. T. (1944). A characteristic dental finding in acrosclerosis and diffuse scleroderma. *American Journal of Orthodontics and Oral Surgery*, 30(1), C25-C29.
8. Jagadish, R., Mehta, D. S., & Jagadish, P. (2012). Oral and periodontal manifestations associated with systemic sclerosis: A case series and review. *Journal of Indian Society of Periodontology*, 16(2), 271.

Severe Periodontal Abnormalities in Systemic Sclerosis (Scleroderma): A Case Report

9. Miller Jr, P. D. (1985). A classification of marginal tissue recession. *The International journal of periodontics & restorative dentistry*, 5(2), 8.
10. LeRoy, E. C., Black, C. M., Fleischmajer, R., Jablonska, S., Krieg, T., Medsger Jr, T. A., ... & Wollheim, F. (1988). Scleroderma (systemic sclerosis): classification, subsets and pathogenesis. *The Journal of rheumatology*, 15(2), 202.
11. Del Rosso, A., & Maddali-Bongi, S. (2014). Oral health in patients with systemic sclerosis. *Rheumatology*, keu179.
12. Mehra, A., & Kumar, S. (2008). Periodontal manifestations in systemic sclerosis: a review. *Dentistry today*, 27(6), 50-52.
13. Auluck, A. (2007). Widening of periodontal ligament space and mandibular resorption in patients with systemic sclerosis. *Dentomaxillofacial Radiology*, 36(7), 441-442.