

Fixed Prosthodontic Rehabilitation in a Wear Patient with Fabry's Disease

Avinash S. Bidra, BDS, MS, FACP

Department of Reconstructive Sciences, University of Connecticut Health Center, Farmington, CT

Keywords

Fabry's disease; medical management; tooth wear; bruxism; diastema; esthetics; fixed rehabilitation; partial group function.

Correspondence

Avinash S. Bidra, Department of Reconstructive Sciences, University of Connecticut Health Center, 263 Farmington Ave., L6078, Farmington, CT 06030. E-mail: avinashbidra@yahoo.com

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Abstract

Fabry's disease is an uncommon X-linked metabolic disorder that leads to abnormal accumulation of glycosphingolipids in the body resulting in a variety of systemic disorders. Few reports have addressed dental findings and management of these patients. This clinical report describes the fixed prosthodontic rehabilitation of an adult male patient with Fabry's disease, who presented with generalized severe wear of the dentition. In addition to numerous systemic morbidities, the patient also presented with intraoral angiokeratomas, telangiactasias, anterior diastemata, bimaxillary prognathism, and other oral findings known to be prominent in these patients. The patient was managed by an interdisciplinary team of dental specialists in close coordination with his nephrologist. The prosthodontic treatment included restorations on all teeth, except mandibular anterior teeth, and the patient was restored with a partial group function scheme of occlusion. At the 3.5-year follow-up appointment, the patient's oral health and integrity of the restorations remained stable. This is the first clinical report describing the prosthodontic management of a patient with Fabry's disease. Unique features related to this patient's fixed prosthodontic treatment include accommodation to complex medical problems, management of maxillary diastemata, and choice of occlusal scheme.

Fabry's disease is a rare X-linked recessive, lysosomal storage metabolic disorder described by Fabry and Anderson in 1898.^{1,2} It is caused by a deficiency of lysosymal enzyme α galactosidase A (AGA).¹ The altered gene is on the mother's X-chromosome, hence her sons have 50% chance of inheriting the disorder and daughters have a 50% chance of being carriers.³ Hemizygous males usually have more pronounced disease manifestations than heterozygous females.² The incidence has been estimated at 1:40,000 to 1:117,000 people worldwide, but absolute numbers are unavailable.¹ A mutation in the gene that controls the AGA enzyme causes insufficient breakdown of lipids that builds up harmful levels of glycosphingolipids in the kidneys, eyes, autonomous nervous system, and cardiovascular system. This results in several clinical signs and symptoms and substantial morbidity and mortality.¹

Systemic manifestations of this disease include cardiovascular disease with susceptibility to stroke at a young age, renal dysfunction, corneal dystrophy, fever from anhidrosis due to sweat gland failure, cutaneous angiokeratomas, and pain in the extremities.^{1,2} Gastrointestinal (GI) disturbance is one of the most common symptoms in these patients.¹ About 19% to 52% of patients report GI symptoms including nausea, vomiting, regurgitation, and abdominal pain. There is no known cure for this disease, and treatment is mainly directed toward management of symptoms such as pain in the extremities (acroparasthesias), hypertension, GI symptoms, hemodialysis (to remove and purify the blood), kidney transplantation for failed kidneys, and enzyme-replacement therapy. Patients without renal transplants often survive into adulthood, but are at increased risk of strokes, heart attack, and renal failure. Patients with renal transplants are reported to survive for more than six decades.¹⁻³

Few articles have described the craniofacial manifestations of patients with this disorder.^{2,4-6} Baccaglini et al² have provided the most comprehensive assessment from their review of 13 patients with Fabry's disease. They reported angiokeratomas on the palpebral and labial cutaneous tissues, intraoral and perioral angiokeratomas, telangiactasias of the labial mucosa and soft palate, cysts/pseudocysts of the maxillary sinus, diastemata in the anterior region, and bimaxillary prognathism. They also noted that xerostomia was present in almost half their patients.²

The dental management of patients with Fabry's disease requires special considerations due to their susceptibility for renal disease, hypertension, and GI disturbances, xerostomia and the numerous medications often taken by these patients. Additional considerations for prosthodontic treatment include management of diastemata for esthetics and optimal occlusal scheme, management of bimaxillary prognathism and the accompanying "gummy smile." Presently, there are no reports in the literature describing the protocol for dental management of such patients. Patients with generalized wear of dentition often present with multiple etiologies that are either mechanical, chemical, or a combination.⁷⁻¹⁰ These etiologies indicate the patients' previous habits, lifestyle, and any underlying medical condition. Several clinical reports have described fixed prosthodontic management of patients with generalized wear of dentition.¹¹⁻¹⁴ The patient's needs, finances, motivation, and time dictate prosthodontic treatment options for patients with generalized tooth wear. Close collaboration with other medical and dental specialists is often necessary.

The purpose of this clinical report is to describe the prosthodontic management of an American College of Prosthodontists Prosthodontic Diagnostic Index¹⁵ (ACP PDI) Class IV dentate patient with Fabry's disease. Unique features related to this patient's prosthodontic treatment include accommodation to complex medical problems, management of anterior diastemata, and choice of occlusal scheme.

Clinical report

History

A 41-year-old Caucasian man presented to the prosthodontist requesting a comprehensive evaluation, in order to improve the esthetics and structure of teeth he recognized as being compromised (Fig 1). He was aware of his complex dental situation and desired a definitive solution. Evaluation of his medical history revealed he had been diagnosed with Fabry's disease during his childhood and presented with multiple conditions associated with his disorder. The patient's medical history included hypertension and bilateral renal failure followed by a renal transplant donated by his sibling. His medical condition resulted in a long history of GI disturbances including nausea, vomiting, and regurgitation. The patient's medications included immunosuppressants, antihypertensives, and drugs for enzyme replacement therapy. Of special consideration was Agalsidase Beta, an enzyme replacement drug administered to him intravenously once every 2 weeks. The patient's dental history revealed multiple teeth with large restorations in all four posterior quadrants. He stated that he had a history of excessive consumption of carbonated beverages many years ago, to stay hydrated during treatment for his renal condition. He also revealed that he was aware of his bruxism habits during the day and was unaware of any nocturnal habits. The patient led an active life and was not in any physical discomfort at the time of presentation.

Findings

Extraoral examination revealed multiple angiokeratomas in the perioral region including the lips (Fig 2). The intraoral soft tissues also showed angiokeratomas and telangiectasias in areas with nonkeratinized mucosa. The gingiva showed minimal inflammation, and the maximum probing depth was 3 mm. The patient did not have objective signs for xerostomia, but a predisposition was not ruled out due to his numerous medications and this characteristic being common in Fabry's disease patients.² The hard tissues revealed diastemata in the maxillary and mandibular anterior region only, a characteristic feature of patients with Fabry's disease.² Generalized severe attrition

and erosion of the dentition was seen on the occlusal surfaces of all posterior teeth and lingual surfaces of maxillary anterior teeth (Fig 3). This was probably due to a combination of his GI condition and history of excessive consumption of carbonated beverages. The facial surfaces of mandibular premolars and lingual surfaces of maxillary premolars showed severe wear. The mandibular incisors showed minimal wear compared to the canines. Multiple amalgam restorations in the posterior region had loss of marginal integrity and were inadequate. All teeth responded positively to vitality testing of the pulp.

There was a 5-mm vertical overlap, with the mandibular incisors contacting the palatal tissues. Clinically, the crowns appeared short due to a combination of wear, altered passive eruption, and large jaw size. The patient presented with Angle's class I molar relationship; the malocclusion was primarily due to anterior spacing and generalized wear that had also resulted in a group function scheme of occlusion. The patient's maximum intercuspal position and centric occlusion were not coincident. Multiple laterotrusive and mediotrusive interferences were noted, which may have aggravated the patient's bruxism.⁷ The patient's caries risk assessment revealed a high score due to the number of restored teeth and his predisposition to xerostomia related to his medical condition.^{16,17} Based on clinical assessment, esthetics, and available freeway space, it was determined there was no loss in the patient's occlusal vertical dimension (OVD). Therefore, the patient met the criteria of category III described by Turner and Missirlian.⁷ The patient also had excessive gingival display due to a combination of altered passive eruption, prognathic maxilla, and hypermobile maxillary lip. Radiographic examination showed acceptable crown-root ratios on all teeth and no bone loss (Fig 4).

Diagnosis and treatment planning

The patient was under close medical supervision by his physician and nephrologist due to his disease and related symptoms. Based on his history and clinical, radiographic, and occlusal findings from mounted diagnostic casts, the patient was diagnosed as an ACP PDI class IV patient.¹⁵ A multidisciplinary team of a prosthodontist, periodontist, orthodontist, and endodontist developed a treatment plan including complete crowns on all teeth except the mandibular incisors. Metal ceramic restorations (JP-1 Alloy; Jensen Dental, North Haven, CT) were planned on all teeth except the molars, which were to receive complete cast crowns made of high noble alloy (JC Alloy; Jensen Dental). Crown-lengthening procedures were planned for all teeth except the mandibular incisors to ameliorate clinical crown length and obtain better resistance form for the preparations. Elective endodontic treatment was planned for the maxillary right second premolar and mandibular left second molar, as the existing restorations were very close to the pulp. The patient refused to have his mandibular canines restored, as he deemed it to be too aggressive, and therefore the treatment plan was modified accordingly.

The treatment plan was presented for approval by the patient's nephrologist who recommended premedication with 2 gm of Amoxicillin, 1 hour prior to all procedures that would involve any bleeding, considering his renal transplant. Additional recommendations included avoidance of



Figure 1 Pretreatment condition of the teeth in maximum intercuspation. Note the presence of diastemata only in the anterior region. Also note the angiokeratomas and telangiactasias of the alveolar and labial mucosa contributing to the reddish appearance.



Figure 2 Angiokeratomas on the lips and perioral region.



Figure 3 (A) Maxillary occlusal view showing generalized wear of teeth. Also note the large size of the maxilla, anterior diastemata, and multiple restored teeth. (B) Mandibular occlusal view showing generalized wear of teeth. Again, note the large size of the mandible, anterior diastemata, and multiple teeth with large restorations.



Figure 5 Teeth with interim prostheses, in centric occlusion. Note acceptable health of gingival tissues.



Figure 6 Finalized tooth preparations. Note "creation" of interdental papilla, once the diastemata were closed and contact was established between the incisors.



B

Figure 7 (A) Maxillary master cast with dies. Note that previous restorations in all posterior teeth were removed and incorporated as part of the tooth preparations. Grooves for resistance forms were placed wherever necessary. (B) Mandibular master cast with dies. Note that previous restorations in all posterior teeth were removed and incorporated as part of the tooth preparations. Grooves for resistance forms were placed wherever necessary.



Figure 4 Pretreatment panoramic radiograph showing acceptable bone levels, crown-root ratios, and large restorations.

epinephrine-containing local anesthetics to prevent changes in hypertension and avoidance of all forms of nonsteroidal antiinflammatory drugs, due to his renal condition. Therefore, acetaminophen was the recommended analgesic of choice. The patient was also prescribed 1.1% sodium fluoride topical dentifrice (Prevident 5000 Plus; Colgate-Palmolive, Morristown, NJ) for use twice daily. The patient was monitored by his nephrologist for systemic health, and his oral health and hygiene were closely monitored during the entire course of treatment.

Preprosthetic treatment

The patient's maxillary incisal edge position was assessed by lip length, display of incisors with lips at repose, patient's age, esthetics, and phonetics.¹⁸ The existing incisal edge positions were acceptable to the patient and clinician (Fig 2). A Luciatype of acrylic resin jig¹⁹ was fabricated over the mandibular incisors on the cast and was adjusted to correspond to a 2-mm increase in OVD at the incisors. Using this jig, a new centric relation record was obtained, and the casts were remounted on a semi-adjustable articulator (Hanau Wide Vue Arcon 183-2; Whip Mix Corp, Louisville, KY) at the planned OVD. Diagnostic waxing was then accomplished on the mounted casts. The patient requested that the diastemata in the maxillary anterior sextant be closed in the final restorations, and the waxing was done accordingly. A partial group function occlusal scheme²¹ was designed for the definitive restorations. After patient approval of the diagnostic waxing, crown-lengthening procedures were accomplished by the periodontist in quadrants, over a series of four appointments. Scalloped vacuum-formed matrices prepared during the diagnostic waxing were used as guides for all crown-lengthening procedures. During the healing period, endodontic treatment on the two planned teeth was accomplished.

Prosthodontic treatment

After 12 weeks of healing from crown lengthening, initial tooth preparations were accomplished, and interim prostheses were cemented. The interim prostheses were fabricated using the diagnostic waxing as a guide. The acrylic resin jig fabricated over the unprepared mandibular incisors acted as an aid to maintain

the desired OVD increase of 2 mm. During subsequent periods of refinement of tooth preparations, a cast dowel and core was fabricated from high noble alloy and cemented on the maxillary right second premolar. The core of the mandibular left second molar was built using silver amalgam material, engaging undercuts in the pulp chamber and protruding 2 mm into the root canals.²⁰ For the remaining tooth preparations, all previous restorations were removed and not replaced. Presence of caries and demineralized dentin were checked with slow-speed round burs and a caries-detecting dye (Sable Seek; Ultradent Products, South Jordan, UT). Special attention was paid to ensure no undercuts existed in the tooth preparations. Grooves were incorporated to augment resistance form due to the short height of the prepared teeth. After refining final tooth preparations, the interim prostheses were relined and cemented accordingly (Fig 5). The interim prostheses had a partial group function occlusal scheme, where guidance was provided by the canines and both premolars in laterotrusive movements.²¹

The patient had the opportunity to experience the interim prostheses for 5 months. This allowed him to thoroughly evaluate his esthetics, function, and comfort and allowed the clinician a satisfactory assessment of the patient's tolerance to the 2-mm OVD increase. Appropriate health of soft tissues was ensured prior to making final impressions (Fig 6). For the impression procedures, the patient was premedicated with 2 gm amoxicillin, and the gingiva was retracted using knitted cords (Ultrapak, Ultradent Products) of varying dimensions, soaked in 21.3% aluminum chloride (Hemodent; Premier Dental, Plymouth Meeting, PA). Thereafter, complete arch impressions of all tooth preparations were made using polyether impression material (Impregum Pentasoft, 3M ESPE, St. Paul, MN) in the maxillary and mandibular arches. The master cast and dies were prepared from these impressions to proceed with fabrication of the restorations (Fig 7). Thereafter, standard fixed prosthodontic principles¹¹⁻¹⁴ were followed, and the definitive restorations were fabricated. The prostheses were tried in the mouth, and minimal adjustments were made for esthetics and occlusion. The definitive restorations were then cemented with resin-modified glass ionomer cement (RelyX Luting Plus; 3M ESPE) (Fig 8). The partial group function occlusal scheme of the interim prostheses was emulated in the definitive restorations as well (Fig 9).

Posttreatment therapy

After final cementation, new diagnostic impressions were made. An occlusal device was fabricated, and the patient was instructed to wear it at night. The patient was given detailed oral hygiene instructions and was advised to continue using the 1.1% sodium fluoride dentifrice for the rest of his life, due to his predisposition for xerostomia. He was also provided with a set of his new diagnostic casts to enable future monitoring of wear, especially on his unrestored mandibular incisors and canines. He was placed on 6-month recall for maintenance of oral health. At a 3.5-year recall, definitive restorations and health of the soft tissues remained stable. The patient remained satisfied with esthetics, function, and comfort of his prostheses (Fig 10). The patient's systemic health remained stable, and he continued his medical treatment with his nephrologist.









С



Figure 8 (A) Maxillary occlusal view showing the final metal ceramic and gold restorations. Note that diastemata have been closed only between central and lateral incisors; (B) Mandibular occlusal view showing the final metal ceramic and gold restorations; (C) Posttreatment panoramic radiograph showing acceptable bone levels, crown-root ratios, and definitive prostheses. Note that a cast dowel-core has been placed on tooth #4 and an amalgam coronal-radicular dowel and core on tooth #18.

Discussion

From a prosthodontic treatment perspective, important factors in the management of this patient with Fabry's disease were: (1) complex medical history, (2) multiple medications taken by the patient, (3) limited choice of anesthetics and analgesics, (4) appointment durations and schedule, (5) management of





Figure 9 (A) Right laterotrusive image shows the developed partial group function scheme. The guidance was provided by the canines and premolars only. (B) Left laterotrusive image shows the developed partial group function scheme. The guidance was provided by the canines and premolars only.



Figure 10 Frontal image taken at final follow-up, showing the posttreatment condition of the teeth in centric occlusion. The health of the gingival tissues is acceptable.

anterior diastemata for esthetics, (6) partial group function occlusal scheme, and (7) patient's predisposition to xerostomia. It is unlikely that Fabry's disease had a direct bearing on the wear of this patient's dentition; however, it is important to note that he had a long history of GI disturbances including vomiting and regurgitation, history of excessive consumption of carbonated beverages to stay hydrated, and bruxism habits related to stress, all of which contributed to tooth wear. It is important that the prosthodontist ensure these patients have been thoroughly evaluated by a gastroenterologist to rule out any premalignancy of the esophageal lining due to chronic vomiting and regurgitation.

The choice of restorative materials for this patient was based on a confluence of factors. Complete gold crowns cast in high noble alloy were used for all molar teeth, due to the patient's history of bruxism. Gold crowns are simple monolithic restorations that require conservative tooth preparations and have proven longevity.^{22,23} They have a lesser chance of fracture, making them an ideal choice of restoration for bruxism patients. The appearance of gold-colored teeth in the posterior region was not of an esthetic concern for this patient. Metal ceramic restorations were used in all the remaining teeth because of their long and successful clinical track record and their ability to appear esthetic with the help of skilled laboratory personnel.24-27 All-ceramic restorations were not considered in the anterior region because of patient's history of bruxism, need for additional tooth reduction, and difficulty of access for potential endodontic treatment in the future, which has been reported to be the primary complication of crowns in fixed prosthodontics.²⁸

As the patient refused to have restorations on his partially worn mandibular canines, it was difficult to obtain a mutually protected articulation with canine guidance, given the positions of these teeth due to anterior diastemata. Addition of composite resin to the mandibular canines was not performed due to degradation of the dentin bond and potential fracture/accelerated wear from the the opposing metal ceramic crowns. Group function is defined as "multiple contact relations between the maxillary and mandibular teeth in lateral movements on the working side whereby simultaneous contact of several teeth acts as a group to distribute occlusal forces."29 In fixed prosthodontics, a complete group function with guidance provided by all posterior teeth may not be the preferred occlusal scheme; this is because forces are higher in the posterior region due to their proximity to the fulcrum of the mandible and hence, an anteriorly directed guidance is more favorable. Therefore, in this patient, a partial group function scheme was adopted, with guidance provided only by canines and both premolars in all laterotrusive movements.²¹

The mandibular incisors did not receive any prosthodontic treatment, as it was determined that the wear on these teeth was not significant enough to justify complete or partial coverage restorations. Furthermore, contact of the mandibular incisors with lingual surfaces of maxillary anterior crowns was successfully achieved in centric occlusion. The patient was cautioned that progressive wear of mandibular canines and incisors would warrant complete coverage restorations in the future.

Though it was clinically determined that the patient had no loss of OVD, a minimal increase of 2 mm at the incisors (about 1 mm in the molar region) was necessary to gain the needed space for restorations. This helped to achieve conservative tooth preparations in the molar regions for the planned gold restorations. Preprosthetic orthodontic treatment was offered as an option for the patient, but he refused it due to the duration of treatment. Therefore, the patient requested that the diastemata be closed through prosthodontic treatment only; however, the diastemata could be successfully closed only between the central and lateral incisors on each side. Closure of the diastema between the lateral incisors and canines could not be accomplished because of the increased space and to avoid violation of esthetic tooth proportions in the definitive restorations.

Summary

This clinical report described the comprehensive fixed prosthodontic rehabilitation of a medically compromised ACP PDI Class IV patient with a rare condition called Fabry's disease. Through an interdisciplinary team of dental specialists in close coordination with his nephrologist, a successful treatment protocol with special considerations was established and executed. Unique features related to this patient's prosthodontic treatment included management of anterior diastemata and use of a partial group function occlusal scheme. At a 3.5-year follow-up appointment, the patient's oral health and integrity of the restorations remained stable. Future prognosis will depend upon the patient's medical health, adherence to oral hygiene regimen, and motivation.

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