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Rehabilitation of a Maxillary Defect Secondary to Recurrent Giant Cell Granuloma

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Abstract

This is a presentation of the treatment history of a young woman with a benign lesion resulting in a large maxillary defect. This patient's complex treatment resulted in a full spectrum of rehabilitation modalities. Her story shows alternative treatment options with the ultimate goal of restoring form, function, and quality of life to a patient with an extensive maxillary defect.

A 23-year old woman was referred for rehabilitation after a third occurrence of Central Giant Cell Granuloma. The lesion that recurred in her anterior mandible was removed, and she was referred for consultation regarding replacement of her mandibular anterior teeth. A comprehensive oral examination revealed that in addition to the mandibular anterior defect, approximately 80% of her hard palate was missing secondary to two previous occurrences of this disease. The prosthesis was in disrepair, and the remaining maxillary teeth had a poor to hopeless prognosis. Although she was referred for a mandibular prosthesis, her maxillary arch became a critical and immediate concern. This patient's oral rehabilitation demonstrates the complexities presented by these challenging defects.

Clinical report

The patient's first lesion was discovered in her maxilla in 1983 at age 6 when some of her permanent teeth failed to erupt. The Central Giant Cell Granuloma was removed without causing an oral–nasal communication. She had a recurrence at age 13 at which time her left and anterior maxilla were removed. The only remaining maxillary teeth were her right first and second molars and an unerupted right third molar. She related that in 1991 a surgical reconstruction using her left fibula, a relatively new procedure at the time, was attempted and failed.

After her surgical reconstruction failed, a transitional obturator was fabricated. All that remained to provide retention for the prosthesis were her maxillary right first and second molars. Two Calcitek (Zimmer, Carlsbad, CA) implants were placed into the remaining palatal bone at the junction of the palatal shelf and alveolar bone to provide additional retention (Fig 1). When first examined, she was wearing her original transitional obturator with a temporary liner. Two wrought-wire clasps and a Hader-type gold clip retained this prosthesis.

This patient was referred for a consultation in 2000 after a mandibular anterior recurrence of a Giant Cell Granuloma was removed and the area grafted. As the patient had accommodated to her ill-fitting maxillary prosthesis, her chief complaint was the missing lower anterior teeth. She presented with a stable transitional removable partial denture replacing her mandibular anterior teeth.

A clinical and radiographic examination of the remaining maxillary teeth revealed moderate horizontal bone loss. The first molar had a fractured palatal root and periapical radiolucencies (Fig 2). The obturator had a temporary resilient liner placed several years earlier and was heavy, unstable, and in poor condition. The size and location of the defect resulted in a type IV classification according to both the Prosthodontic Diagnostic Index and Aramany's classification of maxillary defects. Therefore, notwithstanding the patient's concerns, her maxilla became the focus of our attention.

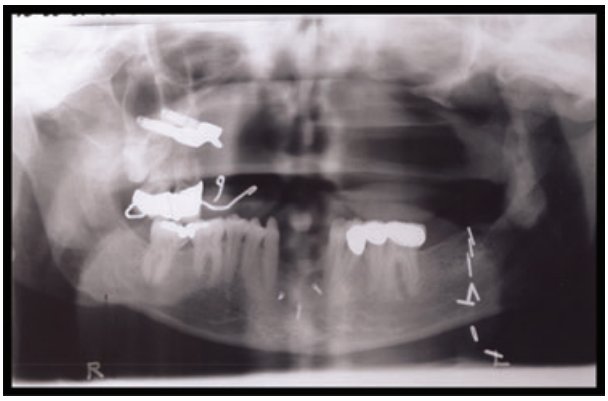

Figure 1 Pretreatment panoramic radiograph.

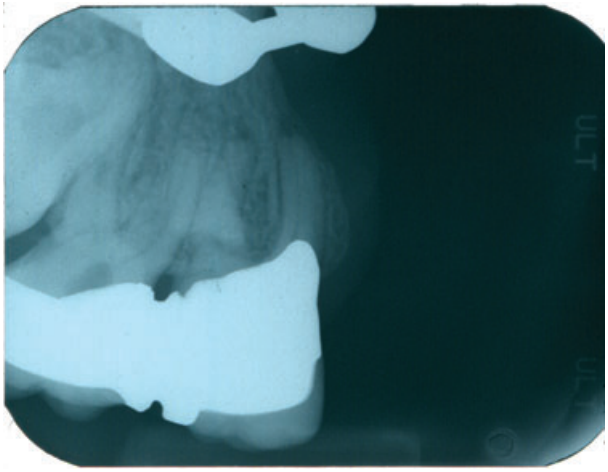

Figure 2 Periapical radiograph of remaining maxillary teeth.

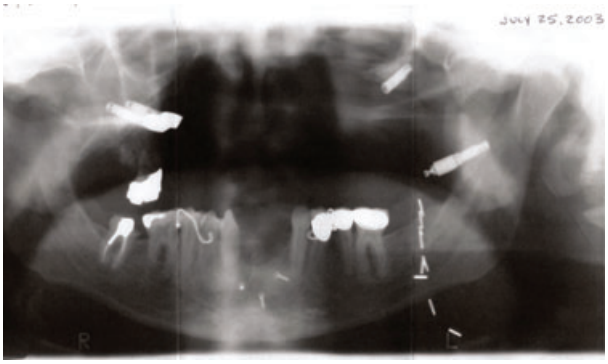

Figure 3 Panoramic radiograph of 2 right Calcitek and 2 left Nobel Biocare implants.

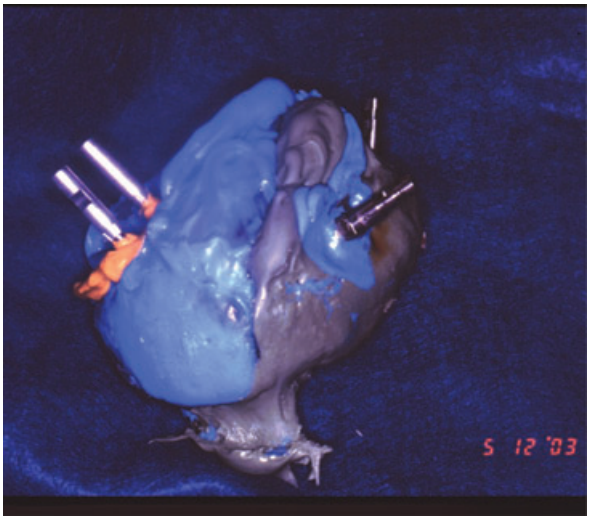

Figure 4 Impression of defect with implant analogs attached.

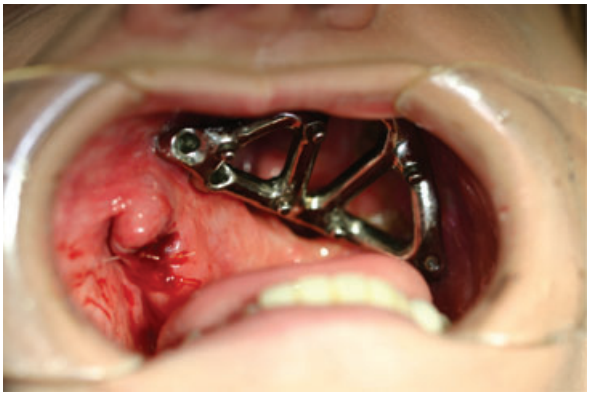

Figure 5 Cross-arch frame immediately postextraction.

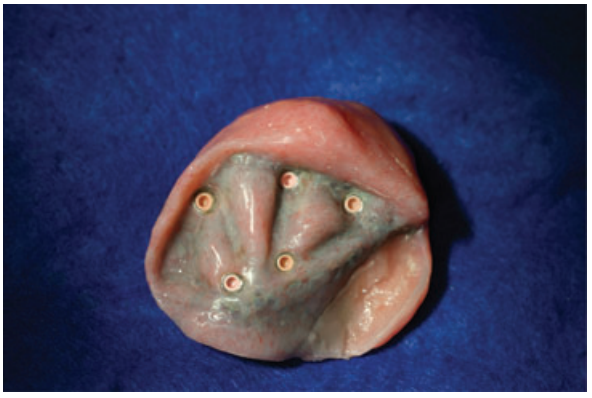

Figure 6 Intaglio surface of completed obturator with 5 OT Cap matrices.

At her consultation, she was informed that the remaining maxillary teeth were hopeless, and we had to explore options for maxillary rehabilitation. Based on her previous experience, the patient refused surgical reconstruction, so zygomatic implants were discussed. She was referred for a 3D CAT scan, and a stereolithographic model was fabricated to facilitate planning of the implants and prosthesis. During this time, the maxillary right second molar became symptomatic and was extracted along with the impacted maxillary right third molar.

In June 2002 implants were placed in the left zygoma and left pterygoid plates (Fig 3). As there was no alveolar bone to pass through, the surgeon was able to place an 18-mm standard Nobel Biocare Implant (Goteborg, Sweden) rather than a

zygomatic fixture. The implants were uncovered in February 2003. A 10-mm standard abutment was placed on the zygomatic implant and just barely reached the surface of the buccal mucosa. Using these two new implants and the two original Calcitek implants, the complex task of fabricating a cross-palatal tissue bar to support a new obturator began. Multiple problems were caused by the instability of the existing obturator, ranging from constant loosening of the zygomatic healing abutment to fracture of the zygomatic abutment screw.

An impression was made, and a master cast fabricated (Fig 4). A jig was then constructed to confirm the accuracy of the cast. The accuracy of the jig was checked clinically, the jig was corrected intraorally, and the cast was then altered to conform to the jig. An accurate master cast was attained, and a cross-arch frame using five OT Cap overdenture patrices (Attachments International, Burlingame, CA) was waxed and cast in a gold alloy. Due to the locations and angles of the implants, the fit of the bar had to be substantiated clinically without the use of radiographic confirmation. In August 2004, this patient's remaining maxillary tooth was extracted. The cross-arch bar was inserted, and the screws torqued (Fig 5). A maxillary obturator with an internal cast metal frame was fabricated, and the five OT Caps were luted intraorally using autopolymerizing acrylic resin at the time of prosthesis delivery (Figs 6, 7). After healing of the extraction site, this area of the prosthesis was relined. The patient was then referred for the placement of three implants in the anterior mandible to address her original chief complaint.

In February 2006 a fistula formed under the patient's left eye. The cross arch bar was removed, revealing a failed ptyergoid implant. The zygomatic implant appeared stable, so it was surmised that movement of the zygomatic abutment secondary to the failure of the pterygoid implant caused the fistula. Though the three remaining maxillary implants appeared stable, considering the existing fistula, we knew that the long-term prognosis of the maxillary restoration was compromised.

Despite this patient's resistance to a surgical reconstruction, it became the only treatment option to ensure a stable definitive prosthesis. After several surgical consultations, the decision was made to have a free fibula flap placed. The iliac crest was considered as a donor site. As it can be easily shaped with osteotomies, it was decided that a superior cosmetic result could be achieved by using the fibula.

The surgical reconstruction was scheduled for May 2006. An acrylic mock-up was made on the stereolithographic model of the patient's skull to approximate the size and shape of the bone graft. This allowed the surgeon and the author to determine the length of fibula required and the location of the osteotomies needed to obtain the proper shape of the bone.

The fibula flap was harvested from the patient's right leg. The fibula was attached to the remaining maxilla with bone plates. Through a submandibular incision, a tunnel was created so the vein and artery from the fibula flap could be anastomosed to the external carotid vessels in the neck. After circulation to the graft was restored, the skin and muscle tissue were positioned to recreate her palate and then sutured in place. Although the incidence of major donor site complications is considered very low, the patient developed a severe infection of the donor site that took almost a year to heal completely. This in turn caused clawed toes on her right foot, requiring surgical correction.

At the end of July 2006, just 3 months after the surgical reconstruction of the maxilla, the flap was de-bulked, and seven osseointegrated implants were placed into the fibular graft (Fig 8). In November 2006, six of the fixtures were uncovered, and the fabrication of a conventional, implant-supported, hybrid-type fixed dental prosthesis was initiated. Due to its angulation, it was decided to leave one implant submerged. A definitive hybrid-type fixed dental prosthesis on the mandibular anterior implants was then completed and inserted in March 2007. Figures 9 and 10 show the definitive prostheses in place. As the cutaneous portion of the flap is full-thickness, there is resulting palatal hair (Fig 11). The patient plans to eventually have laser hair removal, but as of the writing of this report, she has not done so; however, the growth rate and thickness of the palatal hair has decreased with time. The patient remains pleased with the final result as she now has an intact maxilla and palate along with a complete, stable maxillary dentition.

Discussion

The Central Giant Cell Granuloma has an unknown etiology and is a relatively uncommon lesion. It usually occurs in children and has a slight predilection for girls. Especially when it presents at a young age, it can recur and be aggressive. Recurrence rates as high as 72% have been reported.[1] Surgical curettage or resection are the most common therapies, though alternative therapies have been attempted with variable success.[2] No published reports discuss recurrence of these lesions in flap reconstructions.

The maxilla is a complex 3D bone that provides a stable base for occlusion, as well as cosmetic and functional characteristics of the mid-face. Maxillary defects, whether congenital or acquired, can result in a progression of restorative challenges based on the size of the defect. The primary goal of maxillary rehabilitation is the closure of the defect to separate the oral and nasal cavities to allow normal functions of speech and swallowing, as well as to provide a stable base for mastication.

The larger the defect, the less stable an obturator becomes. Instability results in leakage of air and fluids between the oral and nasal cavities, movement of the prosthesis during chewing, and compromised function.[3,4] Stability and retention of a prosthesis become even more difficult when the defect includes portions of the orbital or zygomatic bone. For defects such as these, treatment options other than a traditional obturator should be considered to achieve a reliable and stable restoration.

Before the advent of osseointegration, the only way to achieve retention and limited stability in large defects was by engaging any anatomic undercuts available. When teeth remained, they could be used to assist in the retention of such a prosthesis; however, when confronted with a large defect and an edentulous arch, the sources of retention, support, and stability are nominal at best. Figure 12 demonstrates a prosthesis fabricated for a patient with a total maxillectomy. Flexible, hollowed-out silicone bulbs were used to engage the lateral walls of the maxillary sinuses. Retention and stability of prostheses such as this are poor to adequate. It is also important to

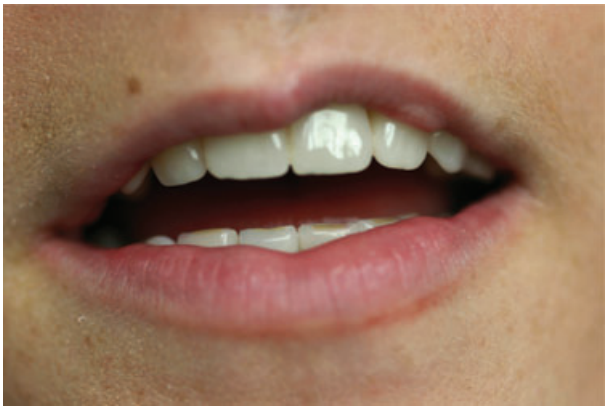

Figure 7 Completed obturator.

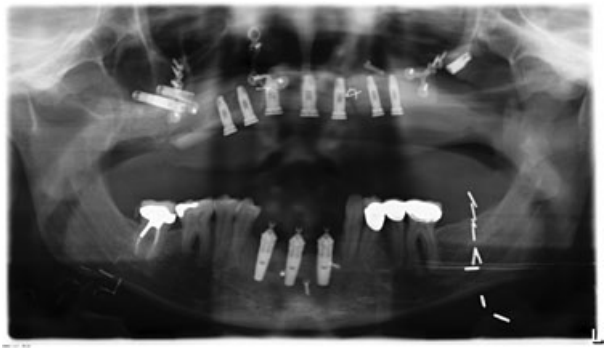

Figure 8 Fibula flap in place with 7 Nobel Biocare implants. Three implants integrated into the mandibular anterior bone graft can be seen. Also note the 2 right Calcitek and left zygomatic implant left in place.

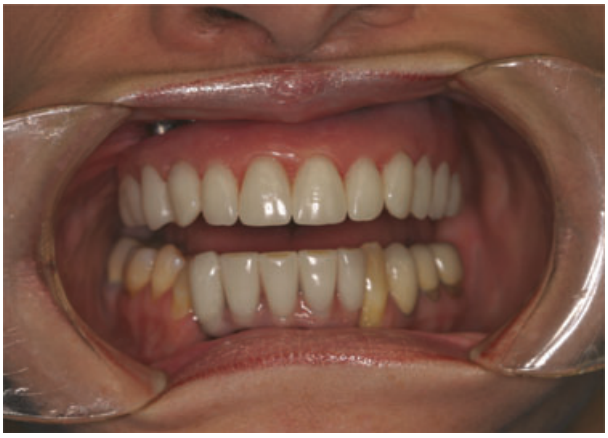

Figure 9 Frontal view of the completed maxillary and mandibular hybrid implant supported prostheses.

understand that the support for this particular prosthesis was provided entirely by nasal mucosa.

Osseointegration brought the ability to achieve a much-improved level of retention, support, and stability to maxillofacial prostheses. Unfortunately implants are not a panacea, as many patients with maxillary defects acquired the defect as a

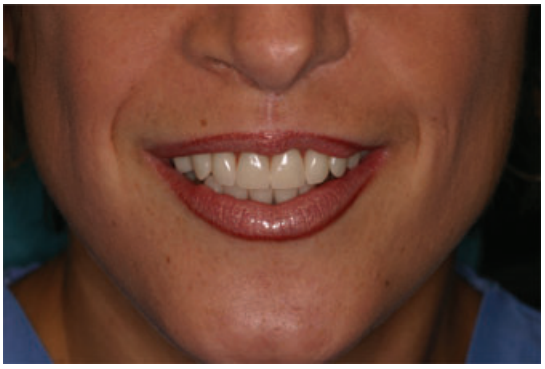

Figure 10 Completed prostheses with a smile.

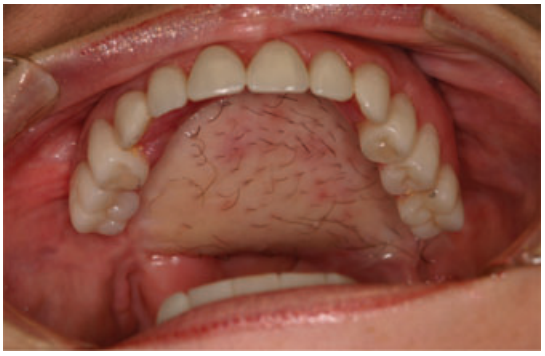

Figure 11 Palatal view of the completed prosthesis.

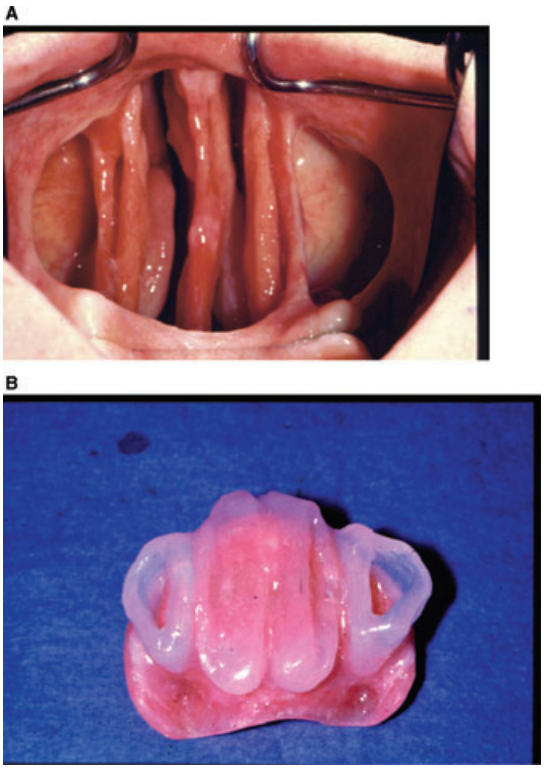

Figure 12 (A) & (B): Obturator for total maxillectomy relying on natural tissue undercuts for retention.

result of surgery to remove a carcinoma. The result is the loss of a great deal of bone and potential implants and often radiation therapy to the area of the defect. With the use of hyperbaric oxygen therapy, it is possible to provide these patients with osseointegrated implants, but the success of implants in these situations are not as predictable as implants placed in healthy bone.

According to the literature, in more than 12 years of follow-up, the zygomatic implant has demonstrated a remarkable success rate of nearly 100%.[5,6] It was developed for use in the atrophic maxilla. The implant extends from the remaining alveolar ridge, through the body of the zygoma, either skirting, or going through, the maxillary sinus. The head of the implant is angled compared to the implant body to allow for an easier connection to a prosthesis. The majority of the published literature on zygomatic fixtures describes its use in conjunction with anterior conventional fixtures. As discussed by Parel et al,[5] cases using zygomatic fixtures require a rigid framework to provide cross-arch stabilization of the implants.

Several published case reports use zygomatic fixtures to provide all of the support where there is a large maxillectomy.[7,8] However, no published studies quantify the success rate of zygomatic implants where there is no connection to traditional implants. When treating patients with large maxillary defects, treatment options to obtain support and retention are limited, and using zygomatic implants may become the only option. The use of these implants in situations where there is no alveolar ridge presents the additional problem of the implant abutments having to pass through relatively thick layers of soft tissue.

An alternative method for palatal rehabilitation is surgical reconstruction. Microvascular free-flap surgery allows the transfer of bone, muscle, and skin along with its own blood supply to recipient sites. The vascularized osteo-cutaneous free flap allows reconstruction of the missing bone, as well as any soft tissue defects. By anastomosing the blood supply of the flap to a vessel at the recipient site, the osteogenic potential of the grafted bone remains intact. This permits placement of the flap, as well as implants in the grafted bone, even in cancer patients who have received radiation therapy.

Two common donor sites are the ileum and scapula. Every donor site that can be used as a source for a free flap has benefits and drawbacks. Each patient and defect must be critically evaluated as to which flap will work best. Although the success rate of these free-flap procedures is high, it is a major surgical procedure that requires at least 1 week of hospitalization. Additional surgical procedures often are required to modify the original flap. Sometimes a second flap is required to obtain complete soft tissue closure of the defect. The donor site always has some degree of negative consequences usually resolved with physical therapy.

Literature reports suggest that the fibula flap has become an increasingly popular choice because it is easy to harvest and is more versatile than the others.[10] The first described use of this flap in the head and neck region was to restore a mandible in 1989.[9] Up to 40 cm of bone can be harvested along with skin, muscle, connective tissue, and a long pedicle vessel. The diameter of this bi-cortical bone is always at least 10 mm and allows easy placement of implants. Reestablishment of the anatomic contours of the maxilla (and the mandible) is facilitated by the excellent blood supply to the fibula that allows multiple osteotomies.[10] The success rate of implants placed into the fibula graft is consistent with the success rate of fixtures placed in any dense bone.[9] The morbidity of the donor site is considered relatively low. A recent study found that 21% of patients had prolonged wound healing caused by infection or wound dehiscence, and abnormalities of gait were observed in 15% of those treated with this flap.[11]

Conclusions

Small maxillary defects, especially in partially dentate patients, can be rehabilitated successfully with removable obturators; however, as the size of a maxillary defect increases, the support, stability, and retention of a prosthesis decreases. An unstable obturator results in impaired speech and difficulties with eating, including nasal regurgitation. Some patients with large maxillectomies are able to receive either conventional or zygomatic implants with the resultant increase in support, stability, and retention. For a variety of reasons, only a small percentage of patients with large maxillectomies are able to receive an osteo-cutaneous flap reconstructing their defect. The success of reconstruction of these defects was unreliable prior to development of free-flap procedures. This technology makes it possible to take patients with severe dental and oral disabilities and predictably provide them with an intact mouth and normal function. This technology enables us to restore a normal quality of life for these patients. We must appreciate, however, what these patients must endure to achieve these results. This reconstruction is a major surgical procedure with possible complications both at the donor and recipient sites.

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