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CLINICAL REPORT

Prosthodontic treatment of a patient with Ewing sarcoma of the left maxillary sinus: A clinical report

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ABSTRACT

This clinical report describes the prosthodontic rehabilitation of a 22-year-old woman with a substantial treatment history of Ewing sarcoma of the left maxillary sinus. The patient was diagnosed with Ewing sarcoma at the age of 7 years and went through chemotherapy, radiation, surgical resection, and free-flap reconstruction, initially without prosthodontic rehabilitation. The patient aged 22 years was referred to the oral oncology clinic at The University of Texas MD Anderson Cancer Center for prosthodontic treatment. The patient's prosthetic rehabilitation with dental implants and a definitive maxillary obturator prosthesis is presented in detail. (J Prosthet Dent 2019;121:698-702)

Ewing sarcoma is a rare, aggressive, and malignant tumor with a small round-cell morphology that affects about 225 children and teens in North America each year.[1] This tumor was first described by James Ewing in 1921 and is thought to arise from undifferentiated osseous mesenchymal cells.[2] Ewing sarcoma accounts for 16% of all primary bone tumors and 5% of all childhood cancers.[1,3] The 5-year relative survival rate for Ewing sarcoma is 77.7% in patients aged less than 1 to 14 years and 61.5% in patients aged 15 to 19 years.[4] Ewing sarcoma usually occurs in the long bones of the upper and lower extremities and less often in the pelvis, ribs, vertebrae, skull, and jawbones.[5] The primary tumor is located in the maxillofacial region in approximately 2% of patients with Ewing sarcoma, most often in the mandible[6-8] and less frequently in the maxilla (about one-eighth as often as the mandible).[9,10] The common signs and symptoms are swelling, paresthesia, and pain. Ewing sarcoma is characterized radiographically by a multilayered "onion skin" or a permeative "moth-eaten" appearance of the affected bone.[3,10]

Reaching a specific diagnosis of Ewing sarcoma is difficult, especially in children, and histopathological and immunohistochemical assessment is paramount.[10,11] The lesion is managed by a multimodality approach that begins with 3 months of neoadjuvant chemotherapy. After chemotherapy, the primary tumor is resected, irradiated, or both, based on its location and extent. Chemotherapy is then continued for up to 1 year.[3]

In this clinical report, the prosthodontic treatment of a 22-year-old woman with Ewing sarcoma is discussed. The patient had been treated with multiple chemotherapy agents, radiation, surgical resection, and free-flap reconstruction, initially without prosthodontic rehabilitation. Secondary free-flap reconstruction and prosthetic rehabilitation with dental implants and a definitive maxillary obturator prosthesis are presented in detail.

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The patient was initially diagnosed with Ewing sarcoma of the left maxillary sinus at the age of 7 years and was

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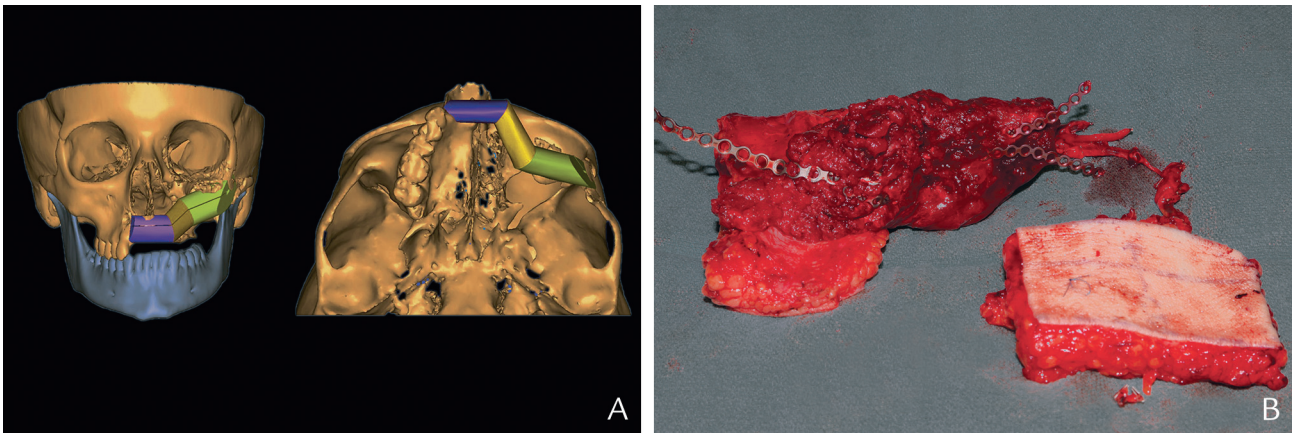

Figure 1. A, Frontal and occlusal view of computer-assisted surgical plan. B, Fibula osteocutaneous free flap and peroneal artery perforator skin flap before being inserted.

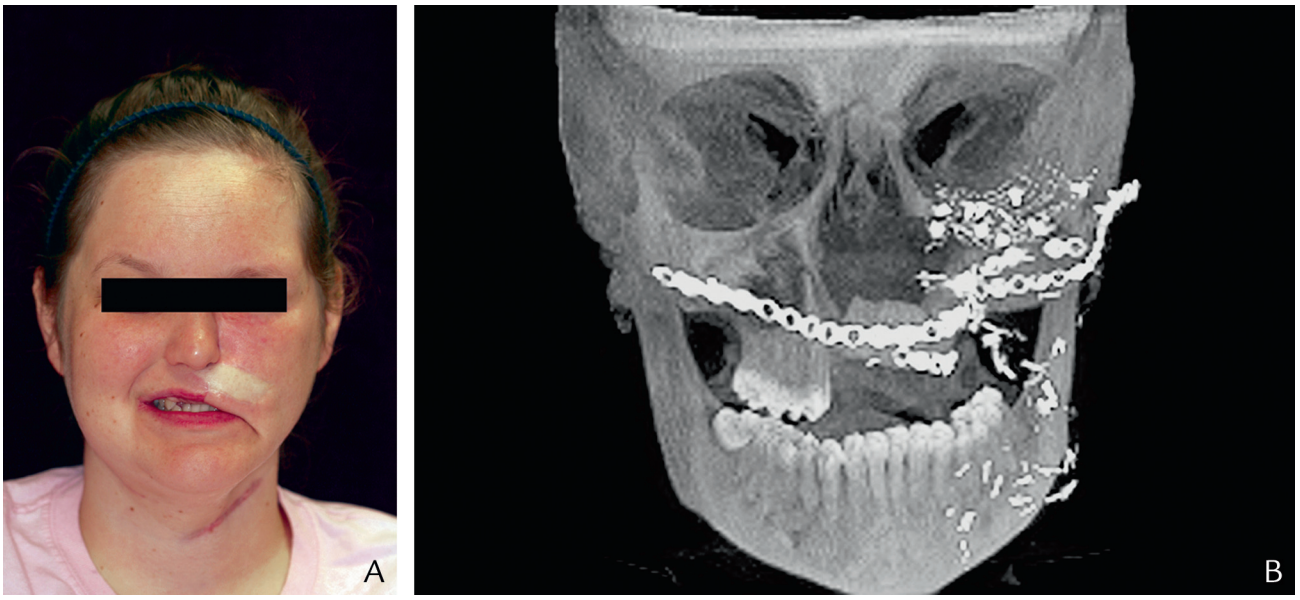

Figure 2. A, Presentation of patient to oral oncology service. B, Radiograph demonstrating fibula osteocutaneous free flap placement. Note additional hardware along previously reconstructed orbital floor.

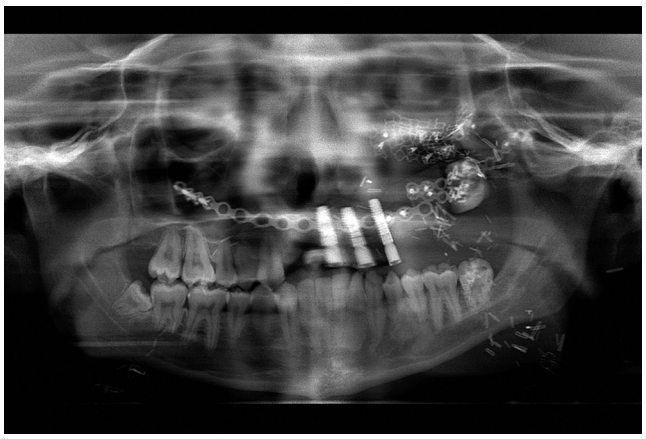

Figure 3. Postoperative panoramic radiograph after placement of dental implants.

treated with chemotherapy consisting of vincristine, doxorubicin, and cyclophosphamide alternating with cycles of etoposide and ifosfamide administered at an outside hospital. Local recurrence was found in the left maxilla 2 years later, and the patient was treated with 3 cycles of salvage chemotherapy (topotecan and cyclophosphamide), followed by 55.8 Gy of radiation therapy. Eighteen months after the completion of radiation therapy, another local recurrence was found. The patient was treated with 2 tumor debunking surgeries and chemotherapy (vincristine, irinotecan, and temozolomide) resulting in 30% shrinkage of the tumor.

At the age of 12 years, the patient was referred to MD Anderson's Head and Neck Surgery Department for further evaluation and treatment recommendations. After consultation with various services at MD Anderson,

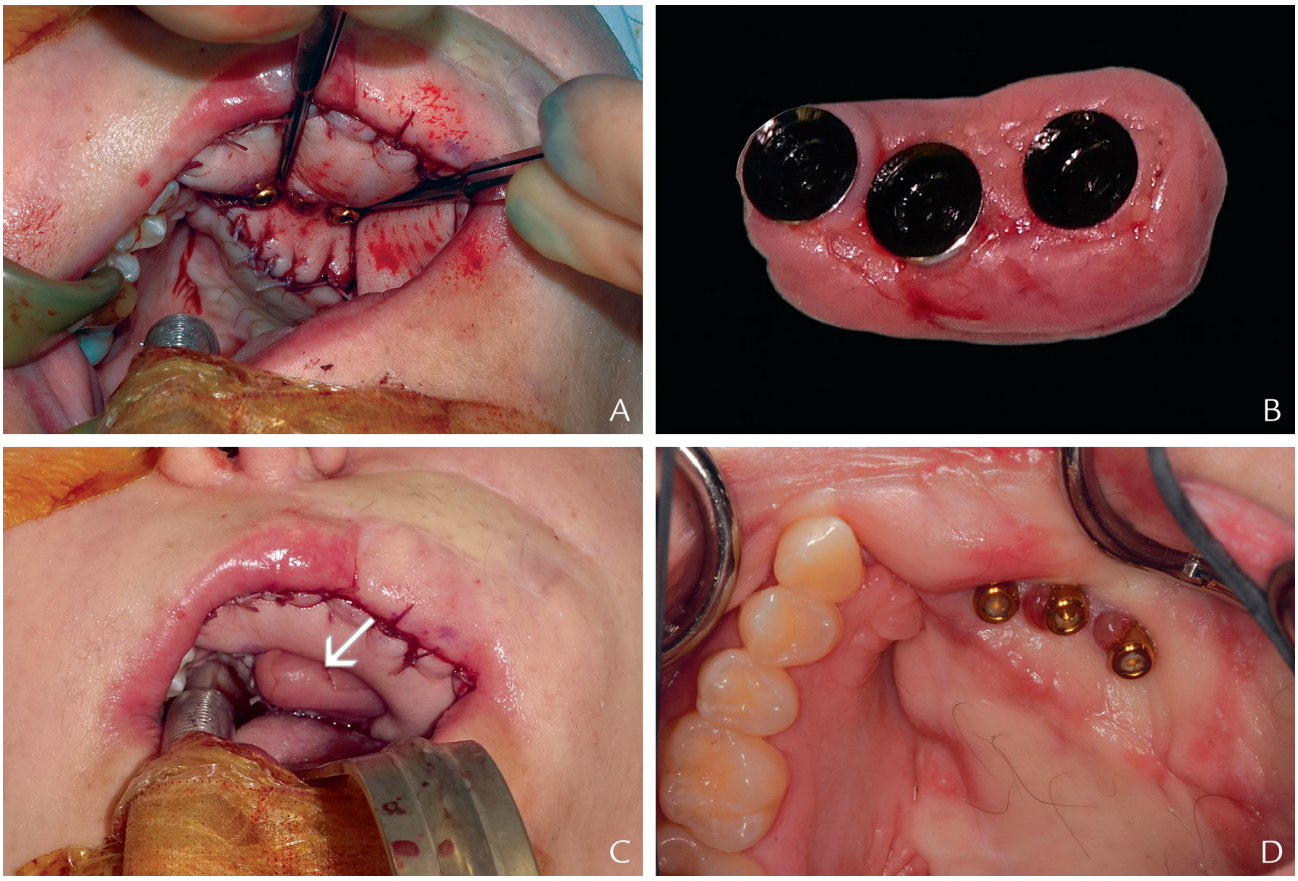

Figure 4. A, Inset of radial forearm fasciocutaneous free flap through Locator abutments. B, Intraoral fabrication of surgical splint. C, Placement of surgical splint to hold flap anteriorly and maintain labial vestibule. D, Three months after treatment showing dental implants in fibula bone with Locator abutments through radial forearm fasciocutaneous free flap.

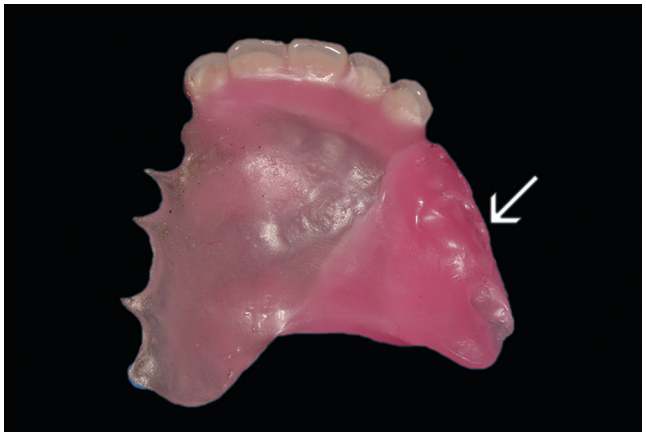

Figure 5. Wax trial tooth arrangement and occlusal generated path (*arrow*).

including head and neck surgery, head and neck medical oncology, plastic surgery, oral oncology, and radiation oncology, the multidisciplinary planning group recommended surgical resection and reconstruction followed by adjuvant chemotherapy. The patient underwent left maxillectomy and reconstruction with a right free anterolateral thigh flap and iliac crest bone graft to the left orbital floor with adjuvant chemotherapy (vincristine, irinotecan, and temozolomide). Prosthodontic rehabilitation was not pursued at that time.

Subsequently, at the age of 20 years, the patient had further reconstructive surgery on the left maxilla with a fibula free flap at an outside hospital. This flap was unsuccessful. At the age of 22 years, the patient returned to MD Anderson's Head and Neck Surgery Department with an orocutaneous fistula and exposed hardware. The multidisciplinary planning group recommended secondary surgical reconstruction of the resected left maxilla. The patient underwent surgical reconstruction of the left and anterior maxilla with a fibula osteocutaneous free flap and reconstruction of the left cheek with a peroneal artery perforator flap (Fig. 1). The reconstructed left maxilla with fibula flap did not receive radiation therapy.

Six months after the surgical reconstruction of the left maxilla and cheek (Fig. 2), the patient presented to the oral oncology clinic at MD Anderson for prosthodontic rehabilitation. A treatment plan was generated to fabricate a definitive maxillary obturator prosthesis with dental implants to replace the missing teeth and to restore lip

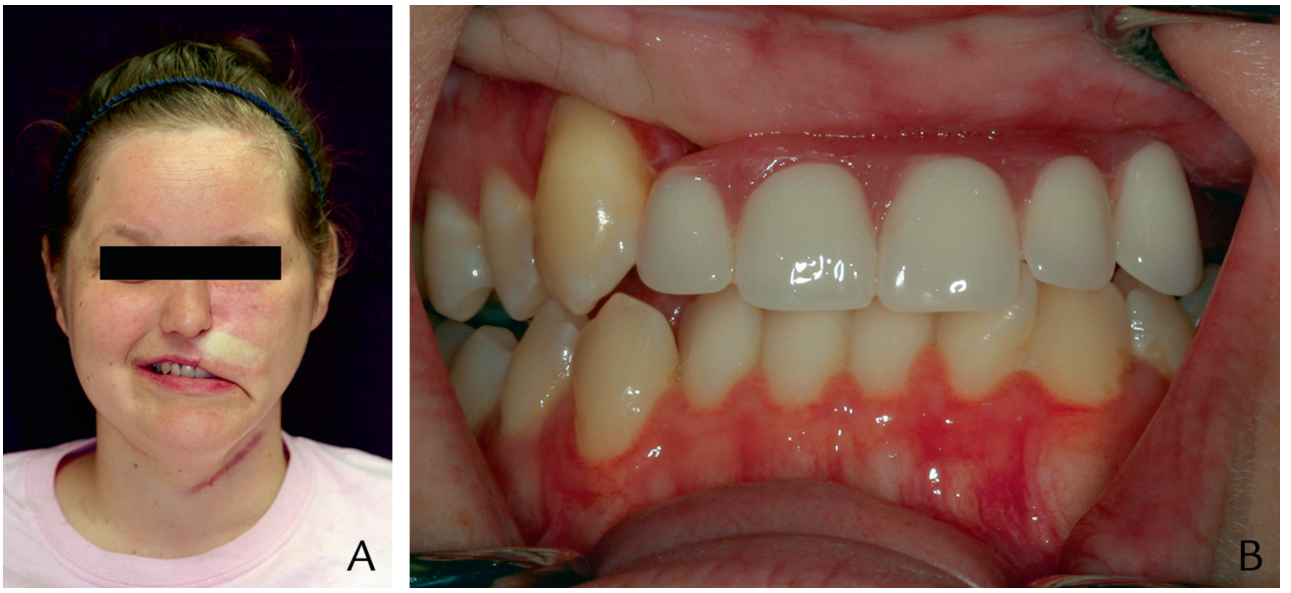

Figure 6. After prosthetic rehabilitation. A, Smile view. B, Frontal view.

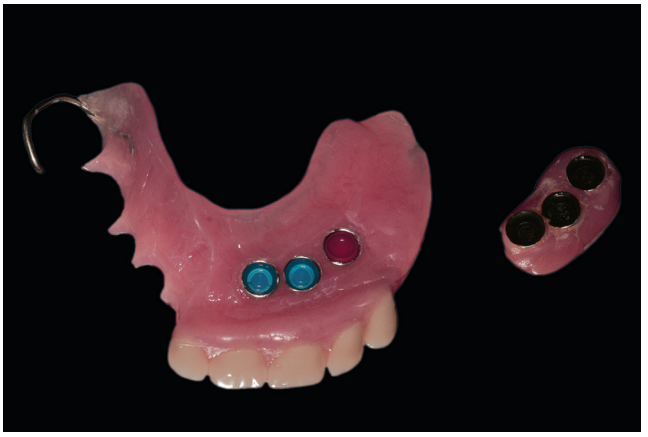

Figure 7. Definitive maxillary obturator and surgical splint.

support, speech, and deglutition. Dental implants were planned to provide retention and stability of the maxillary prosthesis. Two 3.3×12-mm dental implants (Straumann BLT; Straumann USA) and one 3.3×14-mm dental implant were placed into maxillary left anterior sites (Fig. 3). Six months afterward, the 3 implants were uncovered. Cover screws were removed, and 5-mm cuff height Locator abutments (NC Bone Level Locator Abutments; Straumann USA) were placed into the implants. A surgical splint was made in the operating room by luting 3 Locator impression copings together on the abutments with visible light-polymerizing material (Triad; Dentsply Sirona). A radial forearm fasciocutaneous free flap measuring 5×10 cm was used to create an anterior vestibule. Three incisions were made through the flap skin paddle and pushed through the Locator abutments. The surgical splint was then connected to the abutments to hold the flap anteriorly and maintain the labial vestibule (Fig. 4).

Three months after the second-stage implant surgery, diagnostic impressions were made of both the maxillary and mandibular arches with irreversible hydrocolloid impression material (Supergel; Keystone Industries). A maxillary custom tray was fabricated with a visible light-cured material (Fastray LC; Keystone Industries), and border molding of the reconstructed site was done with a green stick modeling plastic impression compound (Kerr Corp). The surgical splint was picked up intraorally in the definitive impression with a light-body polyvinyl siloxane impression material (Aquasil Ultra LV; Dentsply Sirona). The definitive impression was poured with Type IV dental stone (ResinRock; Whip Mix Corp), and the surgical splint was returned to the patient after the definitive cast was produced. A record base was fabricated with heat-polymerizing acrylic resin in the laboratory and adjusted intraorally for proper fit. The left posterior quadrant of the record base was removed because of limited interocclusal space, and baseplate wax was added incrementally to displace the flap until enough space was created for the occlusal generated path (Fig. 5).

The occlusal rim was adjusted according to the esthetics and phonetics. An interocclusal record was made in centric relation by using silicone occlusal registration material (Blu-Mousse; Parkell, Inc), and the casts were mounted on a semiadjustable articulator (96H2 Semi-Adjustable Articulator; Whip Mix Corp) with a facebow transfer. Artificial tooth mold (BlueLine DCL; Ivoclar Vivadent AG) was selected. The mold and esthetic selection was done with the help and approval of the patient. The wax trial tooth arrangement was done on the articulator and verified intraorally with natural dentition and smile line. Lip support and gingival display were achieved by incrementally molding the anterior and superior border

of the flange with dental wax in several visits before heat processing the prosthesis. The incisal position and arrangement of the anterior teeth blended well with the remainder of the dentition (Fig. 6). The patient approved the wax trial tooth arrangement. The prosthesis was invested and processed with heat-polymerizing acrylic resin (Lucitone 199; Dentsply Sirona).

Two blue dual retention and 1 red extended range patrices were inserted. An 18-gauge wrought wire clasp was added for extra retention and as a guide for seating the prosthesis. The patient was instructed to wear the surgical splint at night to prevent soft tissue overgrowth onto the abutments (Fig. 7). The intaglio surface of the maxillary prosthesis had intimate contact with soft tissues and the lingual surfaces of the remaining teeth. The prosthesis was designed in a horseshoe shape for comfort and speech during oral function. The patient's hard palate and the dental implants provided support for the prosthesis. The patient was pleased with the overall esthetics and function of the definitive maxillary obturator prosthesis.

DISCUSSION

For this particular patient, the fibula osteocutaneous free-flap reconstruction of the maxilla presented challenges for prosthetic rehabilitation. These challenges included thick and movable tissue around the implants, constricted restorative space, and reduced bony support for a dental prosthesis. The contracture of the upper lip with high flap adhesion required revision at the time of second-stage implant surgery. A radial forearm fasciocutaneous free flap was used to reconstruct the labial vestibule rather than a skin graft due to the high rate of contracture and skin graft loss in irradiated patients. A potential problem with using a fasciocutaneous flaps is the thickness of the flap in the region of the abutments, making it difficult to accommodate a prosthesis. When used for vestibuloplasty, the radial forearm flap, which is usually the thinnest free flap donor site, is thinned during its dissection and the thinnest part of the flap is placed over the implants. Occasionally, if the radial forearm flap is still too thick, then a secondary debulking procedure is performed 2 to 3 months before prosthesis delivery. For this patient, the flap was adequate, and debulking was not necessary.

Placement of the surgical splint on the abutments played a critical role in favorably shaping the flap in preparation for prosthetic rehabilitation. This also prevented postoperative maxillary lip contraction by providing support for the lip and labial vestibule. The patient was instructed to wear the surgical splint at all times except when cleaning to prevent the flap from overgrowing onto the Locator abutments. It was essential for the patient to learn how to clean efficiently around the abutments and keep the soft tissue healthy. The patient developed gingival inflammatory changes around 2 of the Locator abutments, which is commonly seen with this type of free-flap reconstruction with excess soft tissue around the implants. The inflammation was treated with a professional cleaning and topical steroid ointment (Fluocinonide Cream USP, 0.05%; Taro Pharmaceutical Industries Ltd).

The overall design of the definitive maxillary obturator prosthesis was based on esthetics, oral function, speech, comfort, oral hygiene, and access to surveillance. The removable prosthesis was chosen over a fixed prosthesis for enhanced pink esthetics and modification as the tissues remodeled with time and use. If the strength of the prosthesis were to become an issue, a metal framework could be considered. On the patient's left side, revision of the flap or creation of a vermillion border with a tattoo or lip liners may further enhance the esthetics.

SUMMARY

Ewing sarcoma of the maxilla is an aggressive and debilitating disease that substantially reduces the quality of life of young patients. Some patients require years of chemotherapy, radiation, and surgery before prosthetic rehabilitation is possible. A definitive maxillary obturator prosthesis restores esthetics and oral function for such patients.

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