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CLINICAL REPORT

Customized reconstruction of an extensive mandibular defect: A clinical report

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ABSTRACT

Myoepithelial carcinomas are rare malignant tumors arising from salivary glands. They most commonly involve the parotid and minor salivary glands but may also occur in the submandibular glands. These tumors can become extensive, causing bony expansion and destruction. A 31-year-old man with a large swelling on the left side of the face is presented. Histologic examination of an incisional biopsy confirmed a diagnosis of a myoepithelial carcinoma arising from the left submandibular salivary gland. After tumor resection, the patient's mandible was reconstructed with a customized mandibular framework produced by means of 3-dimensional (3D) laser sintering. This approach significantly reduced cost, advanced surgical procedures, and operating room time, which is of great benefit in a developing country like South Africa. (J Prosthet Dent 2016;116:928-931)

The reconstruction of mandibular defects after resective surgery has always been challenging for surgeons.[1] Several factors need to be considered in the planning of these reconstructions, including the anatomic diversity of the region[2] and the complexity of mandibular movements.[3] Mandibular movements coordinate basic oral functions, including mastication, deglutition, phonetics, and facial muscle tone maintenance. These are important for maintaining life and for social inclusion.[4-7]

Gold standards of treatment for reconstructing segmental defects after resective surgery include advanced microsurgery with fibula-free flaps with costochondral rib and iliac bone grafts.[8,9] These surgeries are rarely possible in developing countries like South Africa because of high costs and shortages of intensive care unit facilities, operating room equipment, and vascular surgeons. Digital reconstruction with 3-dimensional (3D) laser sintering allows the fabrication of an exact titanium replica of the segment to be resected, as a viable treatment alternative.

CLINICAL REPORT

A 31-year-old man presented in August 2013 at the maxillofacial and oral surgery department of a hospital in Kimberley, South Africa (Kimberley Hospital Complex). The patient had had facial swelling on the left side for more than a year before referral (Fig. 1). Several of the patient's teeth had been extracted at other clinics, because these were thought to be contributing to the facial swelling.

Clinical and radiologic examination revealed an expansile mass on the left side of the mandible (Fig. 2), which was causing cortical expansion and spongy (cancellous) bone destruction. An incisional biopsy and subsequent histologic examination of the mass revealed the diagnosis of a myoepithelial carcinoma originating from the left submandibular gland. These are rare malignancies arising in the major or minor salivary glands, and represent 0.2% of all salivary gland tumors.[10]

The treatment indicated for these tumors is complete tumor resection using an extraoral approach with clear surgical margins. This clinical report describes the rehabilitation of an extensive mandibular defect with a customized 3D laser sintered prosthetic framework.

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Figure 1. Frontal view of patient at time of presentation.

Figure 2. Panoramic radiograph at time of presentation.

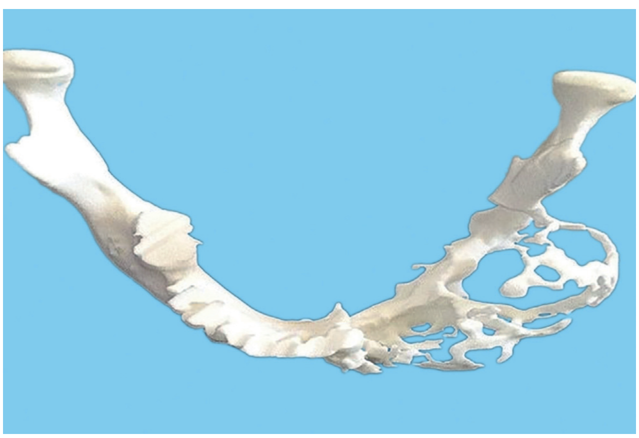

Figure 3. Anatomic model of mandibular hard tissues.

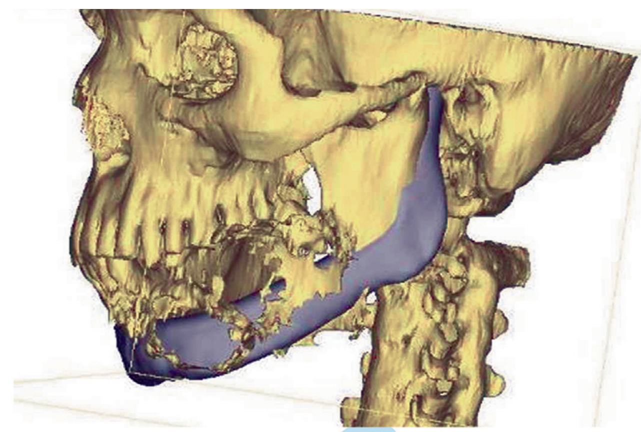

Figure 4. Definitive computer-assisted design of prosthetic framework.

Presurgical planning

Because of the extent of the patient's tumor, an accurate 3D anatomic model had to be fabricated to assist in the surgical planning of the resection and in planning the prosthodontic rehabilitation. New technologies in the form of computer-aided design and computer-aided manufacturing (CAD-CAM) have allowed the accurate replication of 3D anatomic structures in the form of models. This requires data acquisition, data processing, and manufacturing. Data were extracted from computed tomography (CT) scans made on the patient at 1-mm resolution. These scans were imported into a specialized computer software program (Mimics; Materialise NV) for data processing and image segmenting to differentiate between hard and soft tissues. The data obtained as digital imaging and communications in medicine (DICOM) files from the CT scans were transformed into the required stereolithography (STL) files for 3D model printing (additive manufacturing through laser sintering). A polyamide material model (PA2200; EOS GmbH) was produced on a selective laser sintering (SLS) additive manufacturing machine (P385; EOS GmbH) (Fig. 3). This is a Federal Drug Administration-approved material specifically formulated for medical use and is available in powder form (150-μm particle size), which is spread with a roller over the build surface of the laser sintering machine. A piston in the machine's build cylinder moves down 1 object layer thickness at a time while a laser beam traced over the powder surface elevates its temperature to melting point. This fuses the particles close together as a solid mass in the form of a 3D model, as seen in Figure 3.

Prosthesis design and manufacture

A surgical hemimandibulectomy was deemed necessary for complete tumor eradication. This would necessitate extensive reconstruction of the patient's mandible on the left side. The 3D data imported into the software program (Mimics; Materialise NV) was then remanipulated in a secondary software program (3-matic; Materialise NV) to design a prosthetic framework that would be placed into the resected site (Fig. 4). The necessary STL files generated from the software program were transferred directly to the SLS additive manufacturing machine, which produced the framework as per the

Figure 5. Customized prosthetic framework on 3D model.

Figure 6. Postoperative radiograph (posteroanterior skull) with framework in place.

Figure 7. Frontal view of patient at 1-year follow-up with mouth open and closed.

design by sintering medical grade 5 titanium (Ti-6Al-4V) powder of less than 40-μm particle size (Fig. 5). The framework was trimmed, polished, and then sterilized according to the dental implant protocols for the surgical phase of treatment.

Surgical phase

The tumor was resected under general anesthesia, followed by the placement of the prosthetic framework intraoperatively. This framework was secured with 3 bicortical locking screws on the nonresected side of the mandible to secure it in place. The artificial condyle was positioned into the articular surface of the temporal bone to allow for normal ranges of jaw movement postoperatively. A posteroanterior (PA) skull radiograph made immediately postoperatively shows the framework in place (Fig. 6).

Postsurgical prosthetic rehabilitation

The patient achieved excellent results and at 1-year follow-up was clear of tumor. Mouth opening and closing (Fig. 7) was in the normal range, and the mucosa had healed well and showed optimal stability (Fig. 8). The next phase of treatment will include an occlusal rehabilitation of the patient by fabricating a cobalt-chromium removable partial denture with a soft base (Molloplast B; DETAX GmbH) to replace the missing teeth in the mandibular left quadrant.

DISCUSSION

Myoepithelial carcinomas of the submandibular salivary glands are rare malignant tumors. These malignancies often occur in the setting of recurrent benign myoepitheliomas or pleomorphic adenomas.[11] This could

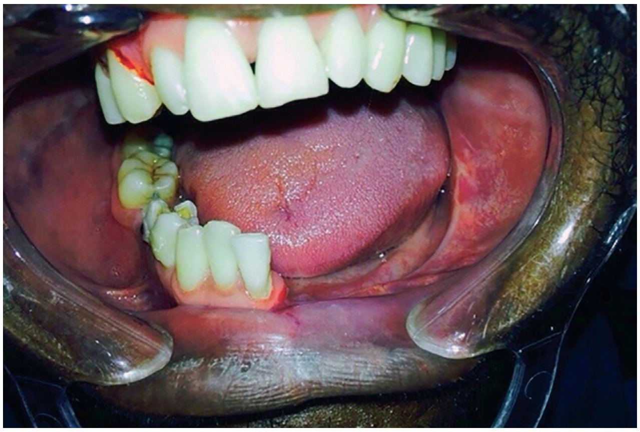

Figure 8. Intraoral view of resected area at 1-year follow-up.

explain the recurrent facial swelling experienced by the patient before referral to the hospital. These tumors have high-grade potential with unpredictable biologic behavior, ranging from localized infiltration to distant metastasis.[11] The 5-year survival rate ranges from 50% to 65%,[11] and thus these malignancies should be completely excised with sufficiently wide tumor-clear margins where possible. Their locally aggressive nature often requires extensive surgical resections for complete tumor excision. When this occurs in the mandible, as in this patient, hemimandibulectomy is indicated. Reconstruction requires advanced microsurgery with fibula free flaps, iliac crest bone grafts to reestablish adequate bone volume, and costochondral rib grafts for reconstruction of the temporomandibular joint. A lack of resources in the public health sector of developing countries such as South Africa results in a shortage of the specialized vascular surgeons required for these reconstructions. Technologic advances in the form of CAD-CAM, 3D printing, and additive manufacturing allows the accurate reproduction of complex anatomic models and design and manufacture of prostheses, which can replace resected segments precisely. A laser sintered titanium framework was fabricated and secured in place with 3 bicortical locking screws on the unaffected right side of the mandible. The prosthetic condylar head was highly polished to allow for articulation on the patient's articular surface of the left temporal bone.

To the best of our knowledge, a prosthetic rehabilitation of this kind has never been carried out in South Africa. The prosthodontic rehabilitation of the occlusion will be carried out in the form of a cobalt-chromium removable partial denture with a soft base to protect the underlying mucosa. The patient is being followed up biannually for close monitoring of tumor recurrence.

Future developments being pursued by the prosthodontist, surgeon, and engineers for these prostheses include coating with hydroxyapatite bone substitute and incorporation of fixture sites for future prosthodontic superstructures, which would allow fixed prosthodontic occlusal rehabilitation in these patients.

CONCLUSION

The use of customized laser sintered prosthetic frameworks for treating large mandibular defects is an innovative approach when advanced microsurgery is not available or feasible. The technology is promising, but further research is needed to evaluate long-term outcomes. Significant knowledge was gained with the treatment of this patient. A team approach is highly recommended when tackling such complex tumors and should involve engineers in the 3D planning, design, and surgical procedures so that they can understand the challenges that surgeons face. 3D printing and titanium laser sintering are valuable in facial reconstruction, in that they reduce surgery time, patient morbidity, and total cost. This technology allows for symmetrical facial reconstruction, which benefits patients tremendously.

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