Customised reconstruction of an extensive mandibular defect: A clinical report.

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ABSTRACT

Myoepithelial carcinomas are rare malignant tumours arising from salivary glands. They most commonly involve the parotid and minor salivary glands, but may also occur in the submandibular glands. These tumours can become extensive, causing bony expansion and destruction. The case of a 31 year old male patient with a large swelling on the left side of the face is presented. Histological examination of an incisional biopsy confirmed a diagnosis of a myoepithelial carcinoma arising from the left submandibular salivary gland. Following tumour resection, the patient's mandible was reconstructed with a customised mandibular framework produced via 3D laser sintering. This approach significantly reduced cost, advanced surgical procedures, and operating theatre time, which is of great benefit in a developing country like South Africa.

Keywords: myoepithelial carcinoma, 3D laser sintering.

INTRODUCTION

Reconstruction of mandibular defects following resective surgery has always been challenging for surgeons.[1] Several factors need to be considered when planning these reconstructions, such as the anatomical diversity of the region[2], and the complexity of mandibular movements.[3] Mandibular movements co-ordinate basic oral functions, including mastication, deglutition, phonetics, and facial muscle tone maintenance. These are vitally important for maintaining life and for social inclusion.[4-7]

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

CASE HISTORY

A 31 year old male patient presented in August 2013 at the Maxillo-facial and oral surgery department of a hospital in Kimberley, South Africa (Kimberley Hospital Complex). The patient complained of facial swelling on the left side which was present for over a year prior to referral (Fig. 1). Several of the patient's teeth had been previously extracted at other clinics, as these were thought to be contributing to the facial swelling.

Clinical and radiological 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 histological 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 tumours.

Treatment indicated for these tumours is complete tumour resection via an extra-oral approach with clear surgical margins. This clinical report describes the rehabilitation of an extensive mandibular defect with a customised 3D laser sintered prosthetic framework.

PROCEDURE

Pre-surgical planning

Due to the extent of the patient's tumour, an accurate 3D anatomical model had to be manufactured to assist in surgical planning of the resection, and in planning the prosthodontic rehabilitation.

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

Figure 2. Panoramic radiograph at time of presentation.

New technologies, in the form of computer aided design and computer aided manufacturing (CAD/CAM) have allowed for accurate replication of 3D anatomical structures in the form of models. This requires data acquisition, data processing, and manufacturing. Data was extracted from computed tomography (CT) scans taken on the patient at 1 mm resolution. These scans were imported into a specialised 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 standard triangulation language (.stl) files for 3D model printing (additive manufacturing via 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 an FDA approved material specifically formulated for medical use and is available in powder form (150 µm particle size), which is spread by a roller over the build surface of the laser sintering machine. A piston in the machine's build cylinder moves down one 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

It was decided that a hemimandibulectomy would have to be performed surgically for complete tumour eradication. This would necessitate an extensive reconstruction of the patient's mandible on the left side. The 3D data imported into the software program (Mimics; Materialise NV) was then re-manipulated in a secondary software program (3-Matic; Materialise NV) for designing 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

Figure 3. Anatomic model of mandibular hard tissues.

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

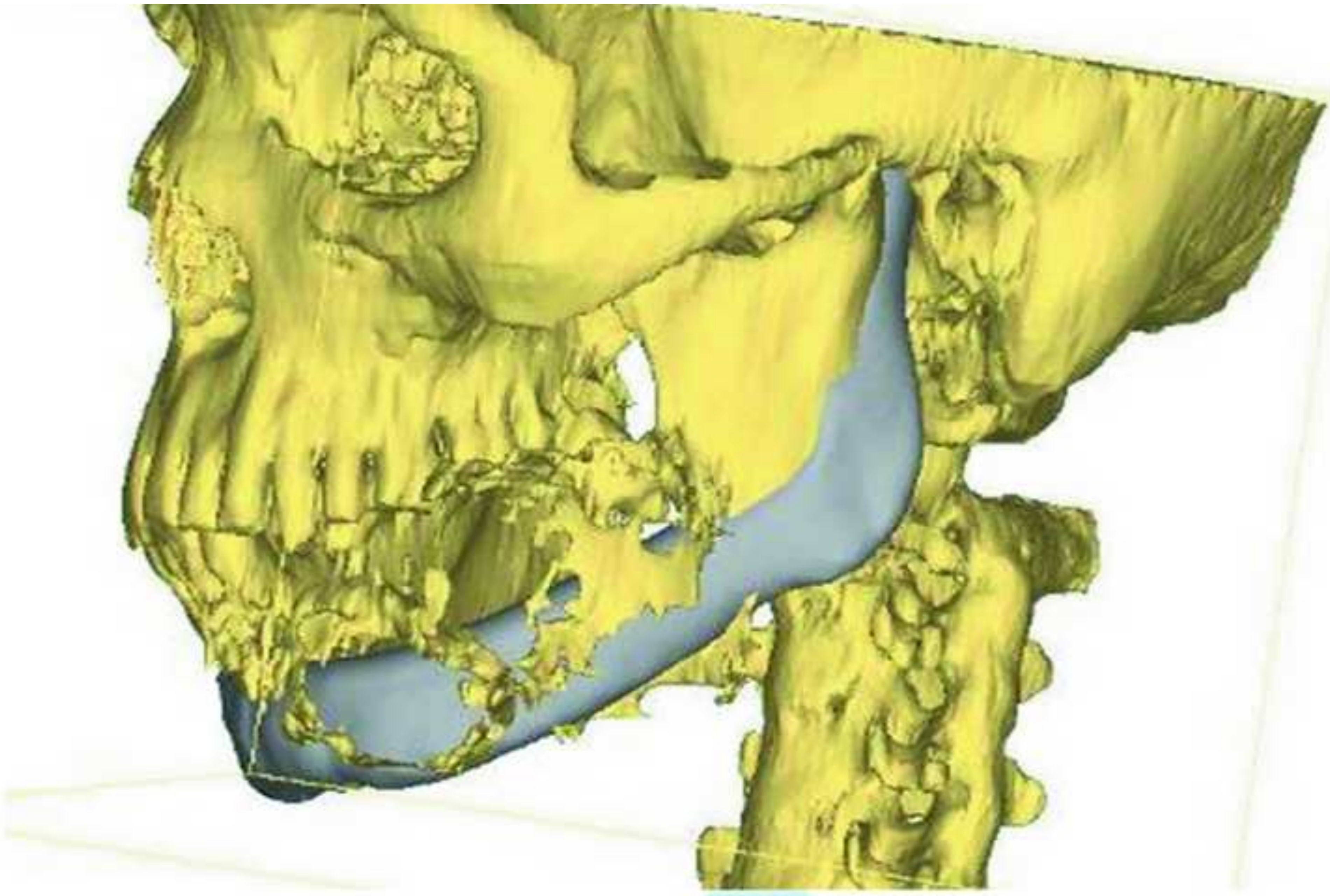

Figure 5. Customized prosthetic framework on 3D model.

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

Surgical phase

The tumour was resected under general anaesthesia, followed by the placement of the prosthetic framework intra-operatively. This framework was secured with three bi-cortical locking screws on the non-resected 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 post-operatively. A postero-anterior skull (PA) radiograph taken immediately post-operatively shows the framework in place (Fig. 6).

Post-surgical prosthodontic rehabilitation

The patient achieved excellent results and at 1 year follow-up is clear of tumour. There is a normal range of mouth opening and closing (Fig. 7), and the mucosa has healed well and shows optimal stability (Fig. 8). The next phase of treatment will include an occlusal rehabilitation of the patient by constructing a partial chrome cobalt denture with a soft base (Molloplast B; Detax GmbH) to replace the missing teeth in the third quadrant.

DISCUSSION

Myoepithelial carcinomas of the submandibular salivary glands are very rare malignant tumours. These malignancies often occur in the setting of recurrent benign myoepitheliomas or pleomorphic adenomas.[10] This could explain the recurrent facial swelling experienced by the patient prior to referral to the hospital. These tumours have high-grade potential with unpredictable biologic behaviour, ranging from localised infiltration to distant metastases.[10] The 5 year survival rate ranges from 50-65%,[10] and thus these malignancies

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.
Figure 7a

Figure 7b

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

should be completely excised with sufficiently wide tumour-clear margins obtained where possible.

Their locally aggressive nature will often require extensive surgical resections for complete tumour excision. When this occurs in the mandible, as with this patient, hemimandibulectomy is indicated. Reconstruction requires advanced microsurgery with fibula free flaps, iliac crest bone grafts to re-establish adequate bone volume, and costochondral rib grafts for the reconstruction of the temporomandibular joint. A lack of resources in the public health sector of developing countries, like South Africa, results in a shortage of specialised vascular surgeons required for these reconstructions. Technological advances in the form of CAD/CAM, 3D printing and additive manufacturing allows for accurate reproduction of complex anatomical models, and design and manufacture of prostheses which can replace resected segments precisely. A laser sintered titanium framework was manufactured and secured in place with three bi-cortical 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.

A prosthetic rehabilitation of this kind had never been carried out in South Africa. The prosthodontic rehabilitation of the occlusion will be carried out in the form of a partial chrome cobalt denture with a soft base to protect the underlying mucosa. This partial denture will be designed and delivered to the patient in the near future. The patient is being followed-up on a regular bi-annually basis for close monitoring of tumour recurrence.

Future research avenues currently being conducted out by the prosthodontist, surgeon, and engineers for these prostheses include coating with hydroxyapatite bone substitute, and the incorporation of fixture sites for future prosthodontic superstructures. This will allow for fixed prosthodontic occlusal rehabilitation in these cases.

CONCLUSION

The use of customised laser sintered prosthetic frameworks for treating large mandibular defects is an innovative approach when advanced microsurgery is not available or feasible. The technology looks promising, however, further research is needed to evaluate the long-term outcomes. Significant knowledge was gained with the treatment of this case. A team approach is highly recommended when tackling such complex cases, which should include the involvement of engineers in 3D planning, designing, and attendance at surgical procedures to understand the challenges surgeons are faced with. 3D printing and titanium laser sintering are very valuable in facial reconstruction, as they reduce surgery time, patient morbidity, and total cost. This technology allows for symmetrical facial reconstruction, which benefits patients tremendously.

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