J Stomatol Oral Maxillofac Surg 121 (2020) 175–178

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Case Report

3D printed titanium prosthesis reconstruction following subtotal maxillectomy for myoepithelial carcinoma – a case report

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ARTICLE INFO

Historique de l'article :
Received 9 March 2019
Accepted 18 March 2019
Available online 25 March 2019

Keywords:
Myoepithelial carcinoma
Maxillary reconstruction
Titanium DMLS frame
midfacial defect
Additive manufacturing (AM)

ABSTRACT

We present the case of a 43 year old male with a myoepithelial carcinoma of the hard palate who underwent a subtotal maxillectomy, resulting in a significant midfacial defect. The defect was successfully reconstructed with a titanium prosthesis using Additive Manufacturing (AM), better known as 3D printing; the process used to manufacture the prosthesis being Direct Metal Laser Sintering (DMLS). A maxillary denture was fitted onto the titanium DMLS frame post-operatively. This method of reconstruction of a large midfacial defect proved to be successful both functionally and cosmetically, and resulted in a good quality of life 3 years post-operatively.

1. Introduction

Myoepithelial carcinoma (MC), also known as malignant myoepithelioma, is a rare salivary gland tumour constituting less than 1% of salivary gland tumours [1,2]. The tumour may arise from the parotid, submandibular or sublingual salivary glands and occasionally the intra-oral minor salivary glands, as was the case in our patient with a tumor of the hard palate [1,2]. Surgery is the mainstay of treatment of these lesions, resulting in variable surgical defects requiring reconstruction, with several options being available for the reconstruction [1,3].

2. Case report

A 43 year old male presented with a 20 year history of a painless slow-growing mass of the palate (Fig. 1). The patient complained of difficulty with swallowing and articulation and started developing malocclusion due to the size of the tumour. On examination, the presence of a large firm mass of the hard and soft palate was noted. Nasal endoscopy revealed a mass on the floor of the nose. A CT scan showed a large homogenous mass involving the maxilla with extensive bony destruction (Fig. 2). Tissue biopsy diagnosed the lesion to be a myoepithelioma.

Surgery with intra-operative reconstruction was planned. Due to the extent and complexity of the planned defect, it was decided to fabricate a titanium Direct Metal Laser Sintering (DMLS) frame for reconstruction. To do this, an anatomical model of the bony tissues was first fabricated as a planning model. The DICOM files from the scanner were converted to STL format using Mimics™ dedicated software (Materialise, Leuven, Belgium) that allowed altering the greyscale values from the DICOM images to differentiate between soft tissue and bone. The region of interest was then marked and calculated as a 3D model, which was sent to the DMLS machine to manufacture the planning model. The Centre for Rapid Prototyping and Manufacturing (CRPM) did the CT segmentation of the skull and produced the 3D model in an EOS P385 Laser Sintering machine (EOS GmbH, Krailling, Germany) in PA 2200 polyamide material at 150 micron layer thickness. From the pre-operative CT scan images, the planned surgical excision borders could be marked, and the defect created by the surgery could accurately be determined. This nylon planning model was sent to the surgical team to be cut where the bone resection was planned (Fig. 3) and served as the template for manufacturing the titanium DMLS frame.

Thereafter, a wax model of the planned titanium frame was made which was reverse-engineered using a Minolta 3D camera and Geomagic® software (3D Systems, Rock Hill, SC, USA). The reverse-engineered geometry was used to identify the boundaries

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https://doi.org/10.1016/j.jormas.2019.03.013
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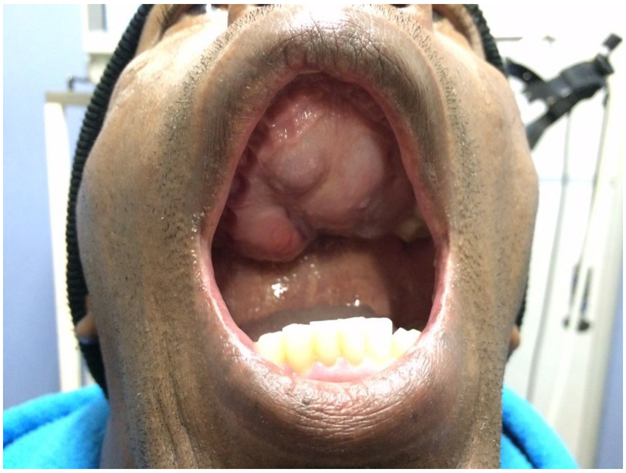

Fig. 1. Pre-operative view of the tumour of the palate.

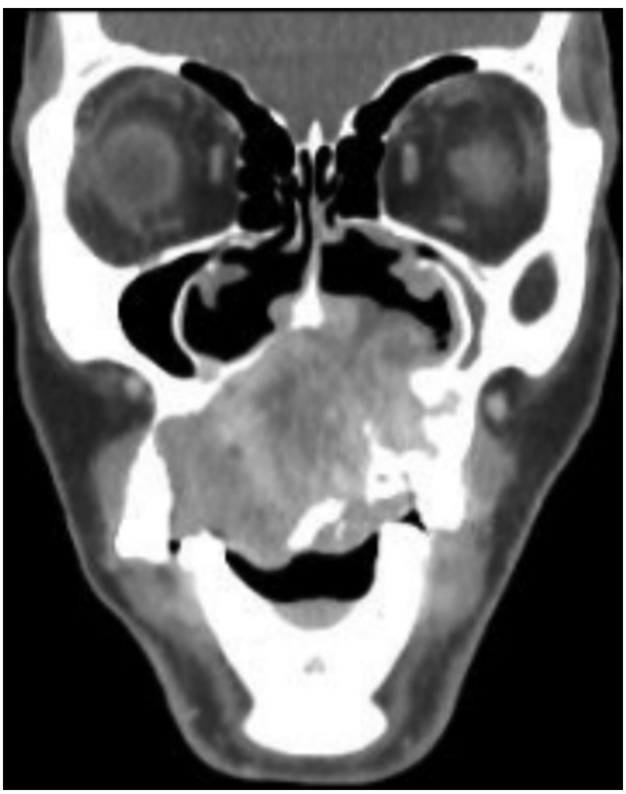

Fig. 2. Coronal CT scan showing the extent of the maxillary tumour with extensive bone destruction/erosion.

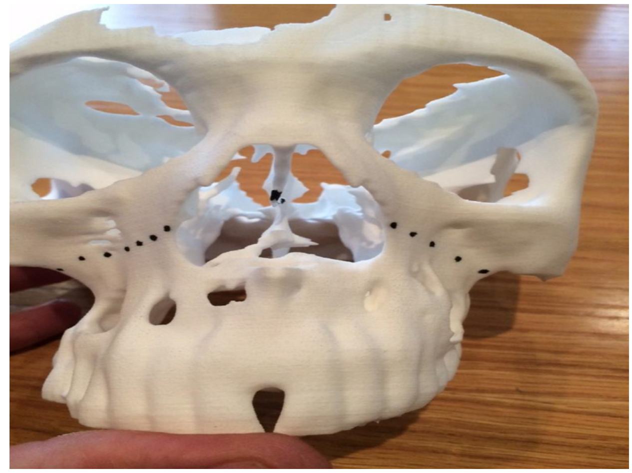

Fig. 3. The anatomical planning model which was additive manufactured from CT images, with dotted lines demonstrating the planned excision plane.

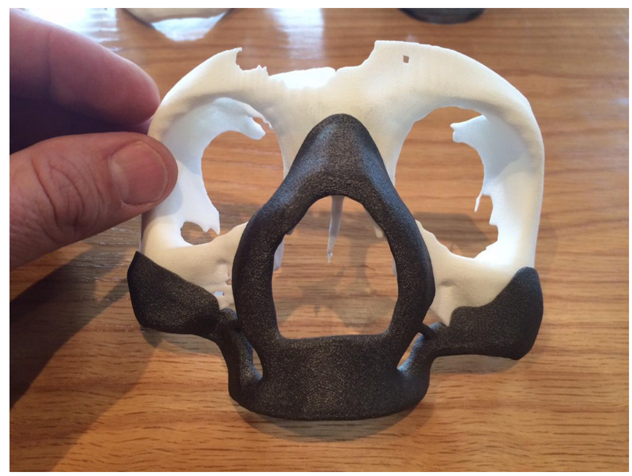

Fig. 4. Demonstration of how the 3D printed nylon prototype fits onto the planning model.

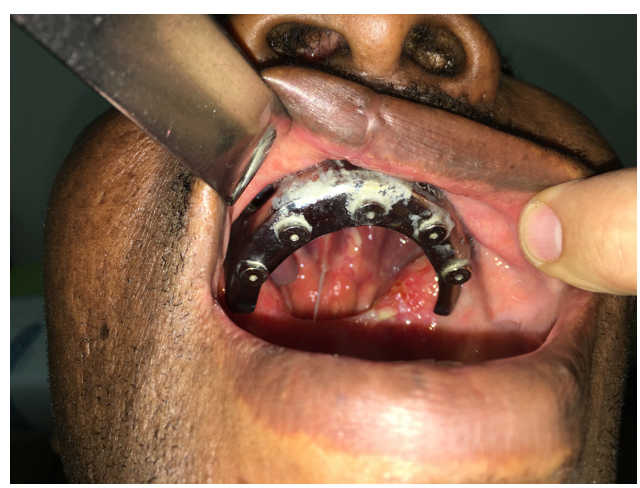

Fig. 5. Post-operative view of the titanium DMLS frame in situ.

of the planned implant. Before designing the titanium implant, a nylon prototype was 3D printed from the reverse-engineered data to fit onto the existing planning model (Fig. 4). This 3D printed customized anatomical model served as the template for printing the titanium prosthesis. The 3D data were imported into 3-MATIC^{TM} software (Materialise, Leuven, Belgium) in order to design the titanium implant. The implant design was transferred to an EOS M280 DMLS machine (EOS GmbH, Krailling, Germany) to manufacture the implant from a biocompatible Ti6Al4V (ELI) powder in 30 micron layers. The support structures were removed and the implant was manually polished. The undersurface of the titanium prosthesis was modified to create an uneven, rough surface to promote osseointegration. Premeasured bone thickness as determined by CT images, dictated the placement of holes in the titanium prosthesis for placement of the cortical screws.

A Dieffenbach-Weber-Fergusson incision was made to create adequate space for placement of the DMLS frame. A subtotal maxillectomy was performed leaving the orbital floor intact

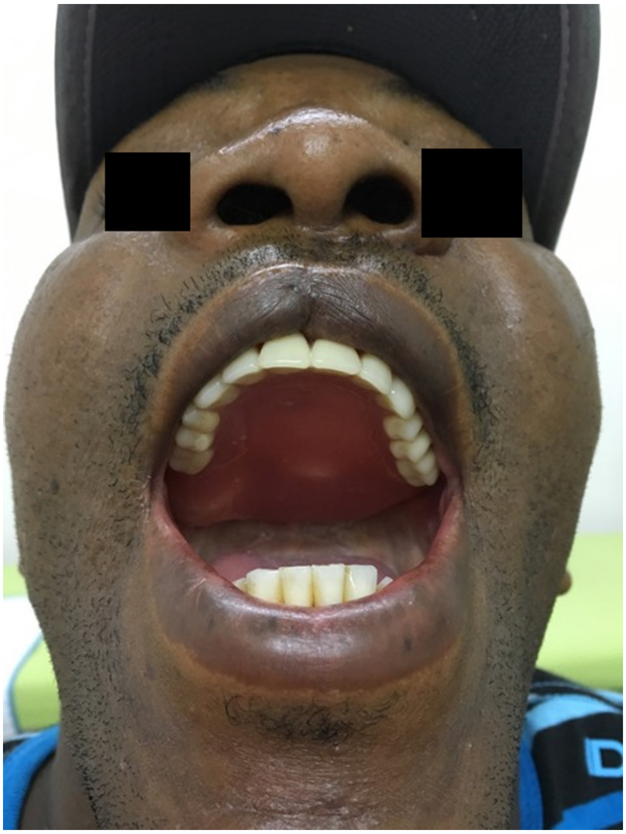

Fig. 6. Post-operative view with removable dental prosthesis fitted onto the titanium plate.

bilaterally, but with complete removal of the hard palate and partial removal of the soft palate. Reconstruction was done with the titanium DMLS frame and secured with cortical screws to the surrounding bone (Fig. 5). A temporary hard palate obturator was placed intra-operatively and sutured to the remaining part of the soft palate. This was removed two weeks post-operatively, followed by placement of maxillary dentures onto the titanium prosthesis (Fig. 6) to complete the reconstruction of the defect. Final histopathology revealed a completely excised myoepithelial carcinoma.

On follow-up after 3 years, the patient remained disease free, reported successful mastication and deglutition with the prosthesis and was satisfied with the cosmetic result. Excellent speech has been achieved post-operatively and the patient is able to independently remove, wash and replace the maxillary dentures.

3. Discussion

Myoepithelial carcinoma (MC) is the malignant counterpart of benign myoepithelioma, the distinction largely being based on the invasiveness of the tumour [4]. MCs are histologically characterized by an infiltrative growth pattern and are almost exclusively composed of myoepithelial cells [4]. They may occur within pre-existing benign lesions such as pleomorphic adenomas or myoepitheliomas (usually low-grade tumours), or may arise de novo (usually high-grade tumours) [1].

MCs may be locally aggressive with a high rate of distant metastases and a tendency for local recurrence [2], but MCs arising in minor salivary glands have a low propensity to metastasize, as these are usually low-grade tumours with low recurrence rates [1,2]. Therefore the treatment of choice for MCs arising from the minor salivary glands is wide local excision with the omission of elective neck dissection [1].

Maxillary defects following surgical resection impair oral functions such as speech, mastication and swallowing, and cause problems with facial aesthetics [5]. There are four types of maxillary defects resulting from surgical resection [3,6]. Type I defects are partial defects where the palate and orbital floor remains intact. Type II defects involve resection of most of the maxilla, but the orbital floor remains intact. Type IIA defects involve less than 50% and type IIB defects more than 50% of the palate. Type III defects result from resection of the entire maxilla with type IIIA excluding and type IIIB including the orbital contents. Type IV defects result from resection of the upper part of the maxilla and orbital contents with the palate remaining largely intact [3,6]. In our patient, the surgery resulted in a type IIB defect.

Several options are available for repair of these defects like free tissue transfer and tissue flaps, dental prostheses supported by zygomatic implants and rapid prototyping. The latter refers to the use of 3D computer-aided design data to manufacture 3D models [6]. It is widely used in the field of dentistry for prosthesis manufacturing [6]. Additive Manufacturing (AM) (as defined by American Society for Testing and Materials (ASTM)) is a process of making a three-dimensional solid object of virtually any shape from a digital model. AM is achieved using an additive process where successive layers of material are laid down in different shapes. Dental prostheses supported by zygomatic implants have also been used to repair these defects, although implant placement may be challenging because of the lack of bony support following surgical resection [5]. There is a higher risk of dental implant failure in irradiated bone including loss of implant, disintegration and peri-implantitis [5]. Ozaki et al reported using two zygomatic implants to support a maxillary prosthesis for oral rehabilitation without maxillary reconstruction for functional and aesthetic rehabilitation in a patient with a maxillary defect after subtotal maxillectomy for a malignant melanoma of the upper gingiva [5]. Peri-implantitis caused by overloading of the implant and implant fracture are potential risks with this technique [5]. Advantages of this technique include no need for maxillary reconstruction, easier prosthesis manufacturing and implant placement as well as decreased surgery time and overall cost [5]. Free tissue flaps and tissue transfer are another alternative for reconstruction of the defect created by the surgical resection. This would have resulted in longer surgical time and the risk of flap failure and subsequent multiple surgeries and increased cost.

The AM technique used in our case proved to be a good option for maxillary reconstruction with potential patient benefit due to decreased theatre time and morbidity. A similar reconstruction method was reported by Fernandes et al for a patient with a peripheral nerve sheath tumour of the anterior maxilla, with a good outcome [6]. Little data on long-term follow-up and complications are currently available because this is a relatively new technique, but our patient had no complications from the implant after 3 years of follow up. Possible complications that may result from titanium plate internal fixation include infection, exposure, pain, cold intolerance, and palpability [7]. Another possible long-term complication is failure of osseo-integration with the risk of extrusion.

4. Conclusion

MCs are locally aggressive tumours and wide local excision is the treatment option of choice. With a large tumour involving the maxilla, wide excision in this area resulted in an extensive mid-facial defect requiring reconstruction. Using a customized titanium DMLS frame and temporary obturator placement, it was possible to repair this extensive defect during the same procedure. This method of reconstruction proved to be successful both functionally and cosmetically, and resulted in a good quality of life three years post-operatively.

Disclosure of interest

The authors declare that they have no competing interest.

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