

## CASE STUDY

# TOTAL MANDIBULAR DEFECT RECONSTRUCTION BY TOTAL TITANIUM PATIENT-SPECIFIC IMPLANT: CLINICAL EFFICACY AND LONG TERM FOLLOW UP. CLINICAL CASE

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The aim of this clinical case is demonstrating the possibility of replacing total defect of the mandible with a patient specific implant and the result of long-term follow up. Literature data on the replacement of total mandibular defects are extremely limited and they are presented by only several clinical cases where various surgical approaches were used. In the available literature, there are two approaches to solving this problem, including the replacement of the jaw with vascularised bone grafts, of which the fibula flap is the most promising, and the implantation of endoprostheses of the jaws, of which patient-specific anatomical endoprostheses made using additive technologies are the most advanced. The concept of using patient-specific endoprostheses of the whole mandible is considered revolutionary because it has a number of significant benefits, including the greatest accuracy in restoring the anatomical shape of the mandible.

One of the unresolved problems associated with the installation of total mandibular endoprostheses is the prosthetic rehabilitation of patients using fixed structures. The analysis of the presented case can be a good tool for the clinician and bioengineer while making the final decision on the treatment method and modality in patients who need an identical option for the repair of a mandibular defect.

Based on CT data, we can conclude that the employed approach, methodology of design and manufacture of patient-specific titanium mandibular endoprosthesis for the total defect demonstrated the sufficient efficacy, which suggests the need for further systematic studies to address this issue.

**KEY WORDS:** Patient specific implant, total mandibular defect, mandible segmentation, mandible reconstruction

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**INTRODUCTION**

Total or subtotal mandibular defects are one of the most pressing and challenging issues in modern maxillofacial surgery. This is due to the fact that the loss of the lower jaw, or a significant part of it inevitably results in profound functional disorders (difficulty chewing, swallowing, breathing and speaking), as well as severe cosmetic deficiencies, facial distortion, social maladaptation, permanent disability, mental disorders and decreased quality of life [1]. The main causes of total and subtotal mandibular defects include tumours [2], osteomyelitis [3], radiation and drug-associated osteonecrosis, congenital malformations such as hemifacial microsomia, the first and second branchial arch syndrome, Pierre Robin sequence and Goldenhar syndrome [4].

In recent decades, a number of approaches have been offered to treat large mandibular defects, which includes those based on the use of 1) free bone grafting procedures [5] or free tissue transferring [6]; 2) conventional reconstructive plates or endoprostheses, based on computer-aided design and manufacturing (CAD/CAM); 3) the combination depending on the clinical situation [7]. Whatever the case, to achieve optimal anatomical and functional outcomes, the authors recommend the use of virtual simulation of surgery and customisation of implants and

fixators (plates, grids, etc.) through model-based bending, milling or additive technologies (DSL) [8].

Until recently, surgeons most widely used microvascular fragmented fibula transferring, usually without reconstruction of the TMJ, forming pseudoarthrosis at the ends of the bone graft [9]. This technique demonstrated acceptable aesthetic and functional outcomes owing to the possibility of dental implants placement and appropriate prosthetic rehabilitation. On the other hand, the authors point to a number of problems and limitations associated with microvascular bone grafting in the area of total mandibular defects, such as a sharp deterioration in the blood supply to the lower third of the face, or traditional recipient vessels may be compromised or absent. Especially, this could be typical defects, caused by drug-associated or radiated osteonecrosis [10].

Numerous prospective and retrospective studies have proven the efficacy of CAD/CAM technology and microvascular tissue transferring for the replacement of mandibular defects [11, 12, 13, 14]. However, the mandibular reconstruction is much more challenging in patients with total mandibular defects [15]. The number of publications concerning total defects is extremely limited, and all of them have data on individual clinical cases. From a topo-

graphic, anatomical and technical point of view, total jaw defects are extremely tough to replace. Due to the very small number of well-documented cases, it is extremely difficult to formulate clear guidelines for the treatment of patients with total mandibular defects. [12]

In 2009, the Dutch company Xilloc together with LayerWise offered a new concept for the treatment of total mandibular defects and an original design for the solid titanium endoprosthesis manufacturing. The developed design was successfully implanted by Belgian professor Jules Poukens in a patient who had needed the lower jaw removal due to bisphosphonate osteonecrosis. In 2011, Professor Jules Poukens and Xilloc CEO Maikel Beerens reported the successful installation of a complete mandibular endoprosthesis in an 83-year-old patient with progressive osteomyelitis [16].

The benefits of this approach include the maximum compliance with the anatomical shape of the patient's lower jaw and an alternative to free tissue transferring in complex cases. At the same time, there is almost no data in the literature on the efficacy of the technique, possible risks and behaviour of the structure with long-term follow up.

Below, we present the clinical case demonstrating our experience of using a total mandibular endoprosthesis in a patient with the total defect, and assessment of the immediate and long-term (more than 2 years) postoperative outcomes.

## CASE REPORT

Aim of this clinical case in demonstrating the possibility of replacing total defect of the mandible with a patient specific implant and the result of long-term follow up.

All performed procedures were in accordance with the ethical standards of the institutional and national research committee, and with the 1964 Helsinki declaration and its later amendments, or comparable ethical standards. The study was approved by the Bogomolets National Medical University Bioethics Committee (Protocol No 126). Written informed consent were obtained from all persons included in the study.

Patient K., 27 years old female, presented to the Centre of Maxillofacial Surgery and Dentistry, Kyiv Regional Clinical Hospital with complaints about an aesthetic defect (sharp deformation of the lower third of the face and upper neck, lower lip, complete secondary adentia) and severe functional deficiency (inability to chew food, difficulty speaking and swallowing, salivation, inability to fully close the lips, sleep apnoea) due to a total mandibular defect. The patient considered the defect to be the cause of deep psycho-emotional disorders (depression, neurasthenia) and a sharp reduction in the social contacts.

The etiology of the defect was associated with the destruction and the mandibular sequestration due to a prolonged diffuse infectious and osteonecrosis due to the systematic use of artisanal derivatives of pervitin [17]. The patient had had this condition for more than 11 years and she underwent 10 surgical interventions (sequestrec-

tomy) with ligation of both external carotid arteries. A long-lasting inflammatory process and previous surgical interventions resulted in marked scarring and atrophy of the soft tissues of the mouth floor and upper neck.

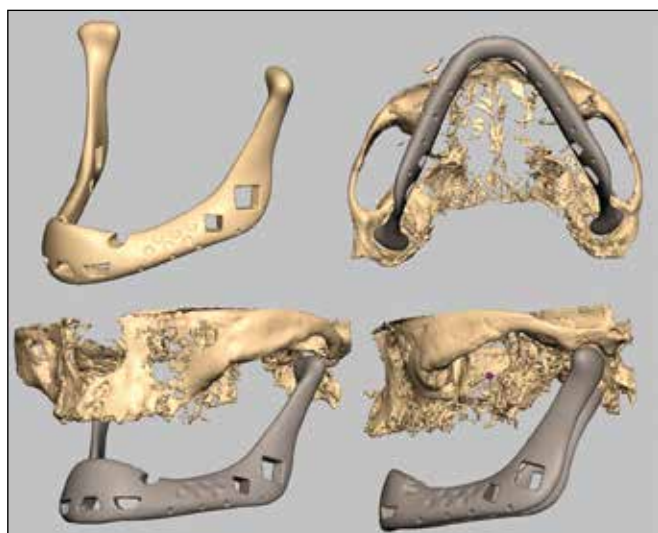
Before starting treatment, the patient had not taken narcotic agents for more than 5 years. The application of the traditional approach with the fibula flap transferring in this case was challenging due to the following factors: the lack of vessels, adequate for anastomosis in the recipient site, the systemic damage to the endothelial lining of vessels caused by pervitin, and the patient's concern about possible complications in the donor site. We took a decision to apply for reconstruction a patient-specific total mandibular endoprosthesis, based on a design offered by Xilloc in 2009.

Before surgery she was evaluated with contrast CT scans and angiography of the head and neck region, as well as both legs. The mandible reconstruction was preceded by an expander placement to create the space for future total patient-specific mandibular endoprosthesis. The expander was installed into anterior part of the floor of the mouth for a period of 4 weeks.

## DESIGN AND MANUFACTURE OF TOTAL MANDIBULAR ENDOPROSTHESIS

CT scans of the patient, as DICOM files (Digital Imaging and Communications in Medicine) were imported into the software environment D2P (DICOM-2-PRINT former Symbionix) ver.1.0.253, 3D Systems, Israel), were CT segmentation was performed with following virtual 3D models' calculation of the facial skull and surrounding soft tissues. To select the parametrically optimal mandible, which was to act as a prototype of the future complete mandibular endoprosthesis, the anthropometrically corresponding mandible was imported, as virtual model following the same segmentation procedures from the CT study of another anonymous patient.

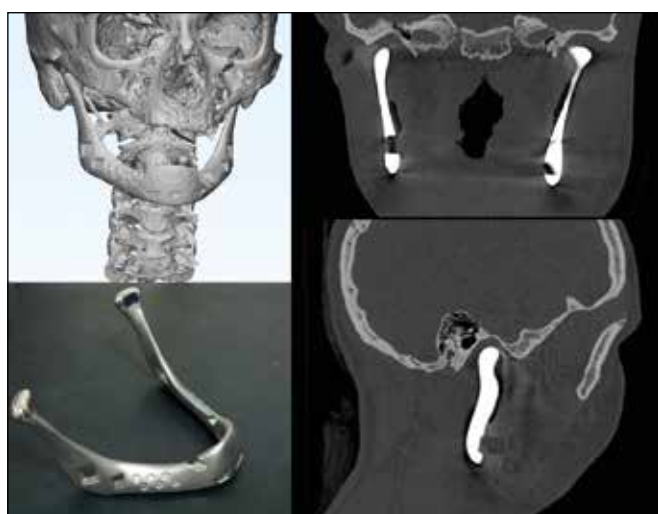
The models, which was created in D2P (3D Systems, USA) as a STL file, were exported to the Geomagic Freeform Plus (version 2016.0 Hotfix 2), where fixing of the model geometry errors was performed. Next, the optimal size and shape of the mandible were determined using the Alignment function and multi-stage verification (profile line, partial cephalometric analysis, soft tissue reproduction, etc.). The "donor" jaw of a certain size was modified in the area of the mandibular rami, creating holes that lightened the construction and served as retention points for attaching the masticatory muscles. Coronal processes were removed from the model, and smaller holes were added along the base of the jaw. The frontal part of the jaw was flattened and rounded, and in the lower third fixation holes were made for a group of muscles of the mouth floor. At the top of the alveolar part of the future implant, there were simulated four fixation holes for fixing the supports of the future prosthetic structure. The volume and linear size of the condyles were also reduced. The three-dimensional model of the created design is shown in Fig. 1.



**Fig. 1.** Virtual design of the full mandible patient specific implant



**Fig. 2.** Patient photos with whole mandible defect: before (upper) and after operation (lower).



**Fig. 3.** Full mandible patient specific implant and CT 3D reconstruction with CT images slices 1 month after surgery.

### MANUFACTURE OF IMPLANTS USING ADDITIVE TECHNOLOGIES

The titanium endoprosthesis was made of Grade 5 (Ti6Al4V) medical titanium, using DMLS technology by European 3D Systems Printing Centre (former Layerwise) Leuven, Belgium. After “printing”, the area of the condyle heads was ground to the mirror surface, while the change in metal thickness did not exceed 30 microns.

### CLINICAL STAGE

Surgery was performed under general anesthesia through the extraoral approaches using previous scars: bilateral submandibular and submental, with preservation of the little skin islands between them. Total endoprosthesis was installed under platysma into created space, the condyle heads of the implant were placed into the articular fossae, the chin was positioned in the midline, masseters, mus-

cles of the tongue and suprahyoid groove were fixed to the endoprosthesis. The patient was prescribed a course of antibacterial therapy and non-steroid analgesics. No complications were seen in the early postoperative period, except some limited mouth opening and foreign body sensation, she required the use of a nasogastric tube for adequate nutrition. The patient assesses the achieved aesthetic outcome as good (Fig. 2).

She refused to have a fixed prosthetic structure installed due to the risk of infection and loss of the structure. Therefore, 4 months following the operation, the patient was installed a removable prosthesis with a soft base. During the entire observation period (27 months), no infectious complications, exposure of the structure or the displacement from the initial position were noted. (Fig. 3)

Literature data on the replacement of total mandibular defects are extremely limited and they are presented by only several clinical cases where various surgical approaches were used. There are currently no clear, scientifically sound guidelines for the replacement of such defects, or the comparison of the clinical efficacy of different techniques. Concurrently, much experience has been gained in the treatment of segmental and subtotal mandibular defects because the availability of preserved parts of the jaw, joint elements and muscle fixation points makes it possible to more accurately determine the desired anatomical shape and position of the lower jaw, as well as more fully restore its functional parameters (the nature of movement in joint, chewing stereotype, etc.) [18]. Therefore, the publication of well-documented case reports of treatment of total mandibular defects with a long observation period and their comparison with previously published data is of considerable interest.

In the available literature, there are two approaches to solving this problem, including the replacement of the jaw with vascularised bone grafts, of which the fibula flap is the most promising, and the implantation of endoprostheses of the jaws, of which patient-specific anatomical endoprostheses made using additive technologies are the

most advanced [19]. The use of the fibula to replace total defects is promising for the following reasons: 1) this method allows restoring the mandible owing to the vital bone tissue that is resistant to infection and integrates well with surrounding soft tissues; 2) the possibility of performing a number of osteotomies allows restoring the mandibular anatomy with high accuracy, as evidenced by satisfactory aesthetic results reported by the authors; 3) the successful installation of dental implants in the site of transplanted fibula demonstrated by van Baar et al. (2018) allowed effective restoring the masticatory function with high-quality prosthetic structures [20]; 4) the possibility of bone grafting together with soft tissues, which can simultaneously replace complex combined defects, 5) the experience of widespread use of this flap in the replacement of smaller defects makes the transplant technique quite predictable in terms of immediate and long-term results. However, the technique has obvious shortcomings and limitations related to the condition of hemodynamics and soft tissues of the donor and recipient sites, systemic diseases of bone tissue and metabolic disorders, risks of flap harvesting, which often cause the patient to refuse surgery and the like.

These problems are most pronounced in the group of patients with drug-associated, toxic and radiation osteonecrosis, which is the most common cause of total mandibular defects. The presented case demonstrates that the transplantation of a free fibula flap was rather challenging and the risk of such an intervention was very significant.

The concept of using patient-specific endoprostheses of the whole mandible is considered revolutionary because it has a number of significant benefits, including the greatest accuracy in restoring the anatomical shape of the mandible, no need for additional trauma to the donor site, a significant reduction in operative time and duration of postoperative rehabilitation, no restrictions associated with the pathology of the great vessels.

The design of the structure in this study broadly reproduced the design offered by Xilloc, but the individual characteristics of the clinical case were taken into account. After the "donor" idealized mandibular model had been imported and cephalometric analysis had been performed, the most optimal size and shape of the mandible were determined. At the next stage, a patient-specific implant was shaped using sequential algorithmic transformations of the prototype with software design tools. Particular attention was paid to the reduced structure weight without loss of the strength and integration of the endoprosthesis with soft tissues.

The proper implant placement, which was radiologically confirmed, improved the aesthetics of the face in the early postoperative period and ensured a significant recovery of lost functions. Concurrently, such an approach raises concerns about the behaviour of the structure during the long-term operation under chewing load. The absence of infectious complications or exposure of the structure in our observation is suggestive of the potential possibility of using such endoprostheses not only as a temporary

solution preceding the final bone grafting, but also as a long-lasting resolution, including lifelong. It should be noted that although the experience of using patient-specific endoprostheses shows a low incidence of complications following the repair of smaller mandibular defects, the incidence of complications in the replacement of total defects may be higher [21].

One of the unresolved problems associated with the installation of total mandibular endoprostheses is the prosthetic rehabilitation of patients using fixed structures. The potential connection of the endoprosthesis with the oral cavity along the transmucosal elements increases the risk of the prosthetic structure loss. In our observation, the patient's refusal of a fixed structure does not allow us to draw any conclusions in this regard. However, our experience shows that the use of removable dentures prostheses is not a functionally complete resolution.

Another controversial issue is the feasibility of two-component patient-specific joint endoprostheses for the replacement of large defects of the jaws with the loss of the mandibular head. Some authors cover this issue [22]. However, they do not reach a consensus, and the results appear to be controversial. The approach employed in our case (with the existing loss of articular discs and components of the joint capsule) is similar to the solution offered by Imad Abu el-Naaj, and it did not provide for the complete restoration of the joint elements. Despite this, the adaptive self-adjusting mechanisms of the masticatory system made it possible to restore the basic parameters of mandibular movements simulating physiological opening and closing of the mouth, as there were no lateral movements (laterotrusion); the inability to contract masticatory muscles and chewing with considerable effort possibly reduced the pressure of the titanium heads on the bone elements of the articular fossa, which was confirmed by the absence of pain and bone remodeling in the site of contact with the endoprosthesis [23].

## CONCLUSIONS

Overall, based on CT data, we can conclude that the employed approach, methodology of design and manufacture of patient-specific titanium mandibular endoprosthesis for the total defect demonstrated the sufficient efficacy, which suggest the need for further systematic studies to address this issue. In such a case, given the lack of CT data of the patient's jaws before the defect, the design of a total mandibular endoprosthesis is a complex bioengineering task that requires special methods and techniques, and the use of total endoprosthesis as such has its drawbacks and advantages. In addition, being very rare, this kind of work requires the involvement of expert-level bioengineers, which brings it closer to the level of exclusive. Therefore, the analysis of the presented case can be a good tool for the clinician and bioengineer while making the final decision on the treatment method and modality in patients who need an identical option for the repair of a mandibular defect.

## REFERENCES

1. Al-Ahmari A., Abouel Nasr E., Moiduddin K. et al. A comparative study on the customized design of mandibular reconstruction plates using finite element method. *Mechanical Engineering*. 2015; 28(1):28-35.
2. Prein J., Hammer B. Reconstruction of extensive anterior defects of the mandible. *Craniomaxillofacial Reconstructive and Corrective Bone Surgery*. Springer, New York, NY. 2002.
3. Neville B.W., Day T.A. Oral cancer and precancerous lesions. *CA Cancer J Clin*. 2002;52(4):195-215.
4. Ashokan C.S., Sreenivasan A., Saraswathy G.K. Et al. Goldenhar syndrome – review with case series. *J Clin Diagn Res*. 2014;8(4):17-9.
5. Foster R.D., Anthony J.P., Sharma A.P. et al. Vascularized bone flaps versus nonvascularized bone grafts for mandibular reconstruction: an outcome analysis of primary bony union and endosseous implant success. *Head Neck*. 1999;21(1):66-71.
6. Schliephake H., Schmelzeisen R., Samii M. et. al. Microvascular reconstruction of the skull base: indications and procedures. *J Oral Maxillofac Surg*. 1999;57(3):233-9.
7. Wilde F., Hanken H., Probst F. et al. Multicenter study on the use of patient-specific CAD/CAM reconstruction plates for mandibular reconstruction. *Comput Assist Radiol Surg*. 2015;10(12):2035-51.
8. Duda T. 3D Metal Printing Technology. *IFAC-PapersOnLine*. 2016;49(29):103-110. DOI: 10.1016/j.ifacol.2016.11.111.
9. Disa J.J., Cordeiro P.G. Mandible reconstruction with microvascular surgery. *Semin Surg Oncol*. 2000;19(3):226-34.
10. Migliorati C.A., Schubert M.M., Peterson D.E. et al. Bisphosphonate-associated osteonecrosis of mandibular and maxillary bone: an emerging oral complication of supportive cancer therapy. *Cancer*. 2005;104(1):83-93.
11. Pogrel M.A., Podlesh S., Anthony J.P. et al.. A comparison of vascularized and nonvascularized bone grafts for reconstruction of mandibular continuity defects. *J Oral Maxillofac Surg*. 1997;55(11):1200-6.
12. Jeremic J.V., Nikolic Z.S., Boricic I.V. et al. Total mandibular reconstruction after resection of rare «honeycomb-like» ameloblastic carcinoma--a case report. *J Craniomaxillofac Surg*. 2010;38(6):465-8.
13. Okada Y., Mataga I., Katagiri M. et. al. An analysis of cervical lymph nodes metastasis in oral squamous cell carcinoma. Relationship between grade of histopathological malignancy and lymph nodes metastasis. *J Oral Maxillofac Surg*. 2003;32(3):284-8.
14. Matros E., Santamaria E., Cordeiro P.G. Standardized templates for shaping the fibula free flap in mandible reconstruction. *J Reconstr Microsurg*. 2013;29(9):619-22.
15. Sato J., Yamazaki Y., Satoh A. et al. Pain may predict poor prognosis in patients with oral squamous cell carcinoma. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2011;111(5):587-92.
16. Nickels L. World's first patient specific jaw implant. *Elsevier Metal Powder Report*. 2002; 67(2):12–14.
17. Poghosyan Y.M., Hakobyan K.A., Poghosyan A.Y. et al. Surgical treatment of jaw osteonecrosis in «Krokodil» drug addicted patients. *J Craniomaxillofac Surg*. 2014;42(8):1639-43.
18. Sirak S.V., Sletov A.A., Shchetinin E.V., et al. Treatment and rehabilitation of patients with subtotal mandibule defects. *Research Journal of Pharmaceutical, Biological and Chemical Sciences*, 2015: 1803–1810.
19. Pellini R., Mercante G., Spriano G. Step-by-step mandibular reconstruction with free fibula flap modelling. *Acta Otorhinolaryngol Ital*. 2012;32(6):405-9.
20. van Baar G.J.C., Forouzanfar T., Liberton N.P.T.J. et al. Accuracy of computer-assisted surgery in mandibular reconstruction: A systematic review. *Oral Oncol*. 2018;84:52-60.
21. Mazzoni S., Marchetti C., Sgarzani R. et al. Prosthetically guided maxillofacial surgery: evaluation of the accuracy of a surgical guide and custom-made bone plate in oncology patients after mandibular reconstruction. *Plast Reconstr Surg*. 2013;131(6):1376-85.
22. Wolford L.M., Mercuri L.G., Schneiderman E.D. et al. Twenty-year follow-up study on a patient-fitted temporomandibular joint prosthesis: the Techmedica/TMJ Concepts device. *J Oral Maxillofac Surg*. 2015;73(5):952-60.
23. Wolford L.M., Mehra P. Custom-made total joint prostheses for temporomandibular joint reconstruction. *Proc (Bayl Univ Med Cent)*. 2000;13(2):135-8.

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**Conflict of interest:**

*The Authors declare no conflict of interest.*

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