Custom made/patient specific alloplastic total temporomandibular joint replacement in immature patient: a case report and short review of literature

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Abstract. – OBJECTIVE: Temporomandibular joint reconstructive surgery in a growing patient represents a challenging situation. Autogenous and alloplastic reconstructive options are being studied in literature; however, there are still some limitations. The objective of this case report is to evaluate a novel custom-made prosthetic system in a 12-year-old TMJ ankylosis patient.

CASE PRESENTATION: The patient had complaints of temporomandibular joint ankylosis and hypoplasia. The patient had already been operated two times with autogenous grafts. Swelling and tumefaction were apparent on the right side of the face. Mouth opening was 1.5 centimeters, with limitations in lateral and protrusive movements of the jaws. Hypertonic muscles and pain upon palpation were registered. There were no signs of luxation, fracture, or traumatic avulsion. After examination, unilateral TMJ ankylosis was apparent on TC scans. Revision surgery was planned with the use of true plastic temporomandibular joint customized prosthesis. The patient underwent a TMJ reconstruction surgery using CADCAM custom-made patient specific prosthesis. The follow up period of this patient was 46 months and showed successful healing with no complications.

CONCLUSIONS: Replacement of TMJ with custom made alloplastic material that is reported can be considered as a safe and useful option for growing young individuals in selected cases.

Key Words: Temporomandibular joint, TMJ, Temporomandibular joint prosthesis, Temporomandibular joint replacement, Alloplastic, TMJR, Growing patient, Pediatric patient, Ankylosis.

Introduction

The reconstruction of the temporomandibular joint (TMJ) represents a challenging situation due to its complex role within the stomatognathic system. TMJ has an essential role in mastication, speech, and swallowing and is subjected to repeated loading/unloading cycles more than any other body joint. Additionally, it supports the respiratory system and is the secondary growth center for the prepubertal mandible1,2.

In the past, due to the complexity of its anatomy and biomechanics, the treatment approach to TMJ problems and pathologies has always been highly conservative. Currently, joint reconstruction is considered to be the only effective treatment in some selected cases1-5.

The accepted indications of the temporomandibular joint that require TMJ replacement surgeries include non-repairable condylar fractures, avascular necrosis, congenital pathologies, bone ankylosis, idiopathic condylar resorption, neoplasms that require extensive resection, patients with previous failed interventions on the temporomandibular joint and severe degenerative joint diseases, such as osteoarthritis6.

The most common need for urgent TMJ reconstruction in growing patients is usually due to
Custom made/patient specific alloplastic total TMJ replacement in immature patient

managing ankylosis or trauma causing cessation of growth\(^6\). The only treatment option for TMJ ankylosis is surgical with or without condylar reconstruction, and various autogenous and alloplastic grafts are available for condylar reconstruction after freeing the ankylotic mass\(^4,6-12\). There is a certain number of scientific papers in literature about autogenous and alloplastic reconstruction options. As a result, these studies\(^13,14\) report improved function and pain scores, but found that autogenous grafts (including the costochondral group) require more revisions. Currently, alloplastic total joint replacement is considered to be the gold standard in reconstruction of the irreparably damaged adult TMJ and there are several custom-made or stock alloplastic options\(^4,6-9,15-17\). The use of alloplastic patient-fitted or stock prostheses has many advantages, such as adaptability, immediate availability, no donor-site morbidity, and reduced operative time. However, surgical experience is required to manage anatomical variability and post-surgical limitations to anterior-inferior movements of the mandible are seen in these patients\(^4,6-9,16,17\).

As an alternative, custom-made prostheses are patient-specific and offer benefits such as wide range of anterior-inferior movements of the mandible and they are anatomically stable. Nevertheless, they have relatively higher costs than stock prostheses and manufacturing time is about 8-12 weeks\(^4,6-9,16,17\).

The primary objective of this case study is to report and evaluate a novel custom-made prosthetic system in an adolescent TMJ ankylosis patient. For this purpose, the patient underwent a TMJ reconstruction surgery using CAD/CAM custom-made patient-specific TMJ prosthesis, and post-operative joint function and pain at 6/12 months were clinically evaluated.

Case Presentation

A 12-year-old Caucasian male patient referred to the Maxillo-facial Department at the “Università degli Studi di Milano – Policlinico di Milano”, Italy, with complaints of TMJ problems. The patient was suffering from swelling and tumefaction on the right side of the face. Upon palpation of the soft tissues on the right side of the face, the osteosynthesis screws on the mandibular angle of the patient could be felt by hand. Mouth opening was recorded as 1.5 cm, with limitations in lateral and protrusive movements of the jaws. Hypertonic muscles and pain were apparent upon palpation on the right side of TMJ. There were not any signs of luxation, fracture, or traumatic avulsion. After clinical and tomography evaluations, it was obvious that the patient was suffering from TMJ ankylosis and hypoplasia TMJ on right side, because of post-partum (after birth) complications. Mimic muscles of the face were normal without any deficit of the nerves or vein. Soft tissues were normal.

Anamnesis of the Patient

The patient had been initially operated about 8 years ago (when the patient was 4 years old) for removal of the ankylosis block and grafting with autologous adipose tissue.

The second intervention was performed after 4 years from the first surgery and included a temporomandibular condylectomy surgery and reconstruction with medial Costa.

After about 4 years from the second revision surgery, the patient referred to the Hospital with TMJ problems. A careful examination of the patient with TC scans confirmed TMJ ankylosis on the right side of TMJ.

Revision surgery was planned with the use of true plastic temporomandibular joint-customized prosthesis under general anesthesia on the right TMJ of the patient.

The surgery was performed with the use of surgical guides produced through three-dimensional (3D) virtual planning methods. The resection of the ankylotic block and reconstruction of the TMJ with a custom-made prosthesis was planned and performed with the use of Mimics Materialise software (Leuven, Belgium). A written informed consent form for the treatment in compliance with the principles of Declaration of Helsinki was provided from the patient before the surgery for the treatment. Additional informed consent was obtained from the patient and the parents for using data including photos for scientific purposes.

Preoperative planning was done by processing DICOM (Digital Imaging and Communications in Medicine) files \(via\) a web-based service with the support of medical engineer (SINTAC, Trento). Planning including resection and reconstruction of ankylosis block and fabrication of the custom made TMJ prosthesis were simulated on the 3D virtual models (SLT files). Dimensions of native mandible was matched with that of new TMJ by an osteotomy through cutting guide to
remove excess osseous tissue. After the final validation, the patient-specific surgical cutting
guides with SLT model and custom made TMJ prosthesis were created within 10 working days.
Figures 1-5 show pre-planning of the patient for surgical treatment.

**Pre-Operative Medications**
Valerian tablets were prescribed for insomnia and psychological stress (taken before sleep). As
a note, the patient had a suspect of allergy against pesto sauce. Two weeks before the surgery, the
patient had panic attack; however, did not receive any additional pharmacological medications.

**The Technical Material Characteristics of Custom-Made System TMJ Prosthesis**
- Pit component: the material used is ultra-high
molecular weight polyethylene (UHMWPE);
- Branch/condyle component: the material used was pure titanium (cpTi);
- Screws for fixing: system 2.0 for the fossa and 2.4 for the mandibular branch in titanium.

Prosthetic system: Sintac (Trento, Italy). Synthesis screws used for fixation: De Puy Synthes
(J&J, Raynham, MA, USA).

**Preoperative Planning**
The patient had undergone a maxillofacial CT scan from the vertex to the chin. Using the data
received from CBCT DICOM files, a computerized and then virtual model of the skull was
obtained to carry out the planning of the surgery.
The case was discussed in a web meeting between surgeons and IT technicians, and the
osteotomies were planned. The custom-made prosthesis was virtually designed adapting it perfectly
to the specific anatomical morphology of the patient.
The joint prosthesis and a stereolithographic model of the skull, cutting guides to be used
during surgery (to ensure that the surgical osteotomy exactly matched to the one virtually
programmed) were then fabricated in the laboratories of the SINTAC S.R.L. Biomedical Engi-
neering (Trento, Italy).

**Surgery: Total TMJ Replacement (TMJ TJR) (Custom-Made)**
Under general anesthesia and after tracheoto-
my, the operation started with a cutaneous pre-
auricular incision with 45° temporal extension.
The incision continued at the right submandibular
site, about 2 cm away from the lower mandibular
border, deepening underneath the platysma mus-
cle, after identification and preservation of the
marginal mandibular nerve, deepening up to the
bony plane of the mandibular angle. The marginal
mandibular branch of the facial nerve passed for-
ward beneath the platysma and depressor anguli
oris muscle, supplying the muscles of the lower
lip and chin, and communicating with the mental
branch of the inferior alveolar nerve. Blunt dis-
section and elevation of subperiosteal detachment
and skeletonization of the angle and mandibular
branch were performed. Skeletonization of the
glenoid fossa of the temporal and elimination
of all residual soft tissues posteriorly, anteriorly,
and medially, were also performed. This step was

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**Figure 1.** A, The pre-operative situation of the patient that was reconstructed from the CBCT and the DICOM files. B, Preoperative situation showing position of teeth, nerves, and fixation screws.
Custom made/patient specific alloplastic total TMJ replacement in immature patient

**Figure 2.** A-B, Pre-surgical planning for mandibular resection and cutting guide

**Figure 3.** A-B, Pre-surgical planning showing the planned resection.

**Figure 4.** Pre-operative planning that based on the symmetry of the collateral site.
important to ensure a direct contact between the bony portion of the fossa and the prosthetic fossa. Deep fascial until the lateral portion of the joint capsule was highlighted. Access to the temporomandibular joint was opened and the retro submandibular incision was performed. Dissection by blunt way up to highlight and skeletonization of the branch of the mandible, the coronoid process, and the sigmoid notch were also performed.

Rigid intermaxillary block was placed in occlusion by using IMF (intermaxillary fixation) screws.

After infiltration anesthesia with vasoconstrictor, the right pre-auricular skin incision (on a previous surgical scar) was cranially extended in the temporal area. The surgery continued with the following procedures: elevation of subcutaneous tissues, subperiosteal detachment and skeletonization of the zygomaticus, and dissection of the joint region, to visualize bone and fibrotic tissue and lysis of cicatricial adhesions in correspondence of the right glenoid fossa.

The operation continued with the osteotomy of the mandibular condyle. The horizontal osteotomy with the second cutting guide was passing through the sigmoid notch and the residual residue of the condylar process. Following the removal of the ankylosis block as planned, the condylar custom-made prostheses (after shaving the right mandibular branch/body and stabilization) was positioned and stabilized by means of synthesis screws by using the holes previously created using the cutting guide. After verification of correct prosthetic articulation and occlusion, careful washing was done by irrigation with serum physiologic and surgical wounds were sutured. The rigid intermaxillar block was removed and the functionality and mobility of the new joint was checked (details on control of the condylar position: if it is correct, the condylar component must be centered in the fossa when the patient is in occlusion).

Bimaxillary rigid fixation (BIM) was achieved with elastics positioned between teeth #33, #34 and #23, #24.

**Post-Operative Regimen**

Immediate postoperative X-ray orthopantomography was performed to evaluate the correct positioning of the prostheses. The patient was hospitalized for two days after surgery.

Post-operative prescriptions:
- Amoxicillin + ac. Clavulanic tablet 1 gr, 1 tablet every 8 hours for 7 days starting one day before surgery.
- Ibuprofen 600 mg sachets, 1 sachet every 8 hours for 7 days.
- Pantoprazole 40 mg tablets, 1 tablet per day for 10 days.

**Follow-up Visits**

1st postoperative follow-up took place at 10 days after surgery.

Parameters evaluated:
- Mandibular opening measurement in mms (this was measured as the distance between the upper and lower arch at level incisal in
maximum opening. The measurement was made with a ruler scale graduated in millimeters;
- Painful symptoms in score: at each check-up, the pain perceived by the patient was assessed. The patient himself measured the intensity assisted by the doctor who provided a visual numerical scale (VAS) from 0 to 10 where 0 corresponds to the absence of pain and 10 to the maximum imaginable pain. There decrease of this score between the pre- and post-operative period constituted one of the main end-points.

Reasons for the visit:
- Wound healing control;
- Control of the general clinical situation;
- Removal of skin stitches;
- Control of occlusion;
- The patient was instructed about joint rehabilitation exercises that must be performed for at least two hours daily: opening movements mandibular with slight forcing (the exercises was continued until obtaining a satisfactory mandibular opening of 25/30 mm).

**2nd postoperative follow-up at 1 month after surgery**
Parameters evaluated:
- Mandibular opening measurement in mms;
- Painful symptoms in score (VAS).

Reasons for the visit:
- Wound healing check;
- Control of the general clinical situation;
- Occlusion control;
- Control of the mandibular opening.

**3rd postoperative follow-up at 2 months after surgery**
Parameters evaluated:
- Mandibular opening measurement in mms;
- Painful symptoms in score (VAS).

Reasons for the visit:
- Control of the general clinical situation;
- Assessment of the degree of rehabilitation;
- Articulation and occlusion control;
- CT cone beam request.

**4th postoperative follow-up at 6 months after surgery**
Parameters evaluated:
- Mandibular opening measurement in mms;
- Painful symptoms in score (VAS).

This patient was followed up for 46 months and the condition of the patient is still under control by regular follow-up visits every 12 months with no further post-operative complications registered so far.

**Discussion**

Regardless of the type of prosthesis chosen for the reconstruction of the TMJ, the main objectives of this reconstructive surgery are the improvement of the joint function and the reduction of painful symptoms.

The first experiences with alloplastic reconstruction of the temporomandibular joint date back to the 1980s with the introduction of the Vitek-Kent prostheses; the main material used for these prostheses was Proplast/Teflon. They were the most used prostheses throughout the 80s and early 90s and early P/T studies were very encouraging with a high success rate. However, in the following years at a longer term through clinical check-ups and radiographic follow-up, it was found that many patients were suffering from condylar resorption, malocclusion, severe pain, and proliferative giant cell foreign body reaction with local destruction of bone and soft tissues, which continued even after the removal of the PT prosthesis. Furthermore, there were numerous reports in the literature about complications, including: perforation of the prosthesis, unstable occlusion, lymphadenopathy, severe osteoarthritis, perforation of the middle cranial fossa, headache, pain and a multitude of systemic disorders such as immunological dysfunction and malnutrition. All these led the FDA to discontinue the production and use of temporomandibular joint prostheses in 1993. Since then, alloplastic reconstructions with joint prostheses temporo-mandibular were partially abandoned in favor of autologous tissue reconstructions.

In 1999, TMJ Concepts prostheses were presented in the market as a breakthrough and were approved by the FDA. These prostheses were considered to be an innovation as they were built according to the specific anatomical situation of
the patients (custom-made) and with new materials (chrome-cobalt-titanium for the branch-condyle part and very high molecular weight polyethylene for the pit part glenoid). During the same period, another type of prosthesis (Biomet® prosthesis, Zimmer Biomet, Milan, Italy) also made its way, this time in stock (pre-formed), receiving FDA approval in 20053,4,7,8,19. Currently, TMJ Concepts® (custom-made system, Ventura, CA, USA) and Biomet® (stock and custom-made system, Zimmer Biomet, Milan, Italy) are still two of the most utilized alloplastic reconstruction systems of the temporomandibular joint and both find their indications for use based on the characteristics of the pathology and the patient.

Lotesto et al6, in 2017, assessed the number of alloplastic total temporomandibular joint replacement (TMJ TJR) devices implanted and the complications encountered by members of the American Society of Temporomandibular Joint Surgeons (ASTMJS). For this purpose, a questionnaire was developed using REDCap (Chicago, IL, USA) and an online link was e-mailed four times over a 6-week period (February-March 2015) to all members of the ASTMJS.

According to the results of this work, etiology of TMJ TJR failures include infection, ankylosis, material hypersensitivity, device failure, fixation loosening, and chronic pain or dysfunction6. Poor component fit, dislocation, screw fracture, and significant bleeding was noted as possible intra-operative complications6. According to the opinions of the surgeons on the improvement of future alloplastic TMJ TJR, the majority of respondents felt that improvements in 3D surgical planning and surgeon training are mandatory and improvement in materials and surgical navigation systems would enhance outcomes. Regarding the lifespan of the TMJ TJR devices, the majority of surgeons (94.4%) expected the TMJ TJR devices to last 10 years or more6.

Currently, alloplastic total joint replacement is considered to be the gold standard in reconstruction of the irreparably damaged adult TMJ. The current prostheses in the market have up to 20 years of follow-up with good outcomes5. In cases of children, the reconstruction of the TMJ is usually secondary to ankylosis or trauma or early onset rheumatoid disease that can cause cessation of growth. In the cases of the children/adolescents, it is important when to consider reconstruction. It is critical that the disease process causing the problem is appropriately managed before considering any operation5.

Majority of current reconstructive techniques favor the use of autogenous replacement in children and alloplastic materials in adults5. Classically, pathologic, developmental, and functional disorders affecting the TMJ in children have been reconstructed with autogenous tissues (costochondral grafts or other autogenous bone/cartilage combinations)20-24. In theory, these autogenous grafts have growth potential and can grow with the young patient. However, this unpredictable growth potential can encounter some problems. Long term reports of mandibular growth in children who underwent TMJ reconstruction with autogenous tissues show some possible negative outcomes, such as excessive growth, inability of the graft to adapt to the growth velocity of the new environment, and ankylosis; more recent studies20,25-35 have even questioned the necessity for using a cartilaginous graft to restore and maintain mandibular growth. Other disadvantages of CCGs include the need for a secondary surgical donor site, possible iatrogenic pneumothorax, and increased operation time20.

When autogenous grafts fail to incorporate into the host bone, it grows horizontally rather than vertically, or might become ankylosed. This causes critical problems and most often the patient needs a revision surgery, such as debridement or replacement of the joint, with other types of allografts. Sometimes, the operation is postponed until the child reaches physical maturity, being operated with another autogenous graft and/or orthognathic surgery and/or distraction osteogenesis procedures. During this period, it is inevitable that this patient becomes disabled and encounters several problems, including function and social limitations, which can lead to depression, and very poor quality of life19.

The introduction of alloplastic materials has improved the quality of life for many adult orthopedic and TMJ patients with unsalvageable functional and anatomic joint pathology. According to the literature, most of the surgeons prefer alloplastic TMJ prostheses, except in the growing patient. However, due to the possible complications of autogenous grafts in growing individuals, as stated above, and along with the reported success of alloplastic TMJ prostheses, it seems reasonable to consider the possibility of TMJ reconstruction with alloplastic prostheses in certain pediatric populations5,15,20,36-38.

More recently, due to the encouraging reports from experienced surgeons, the balance seems to be swinging towards alloplastics in older children,
Custom made/patient specific alloplastic total TMJ replacement in immature patient

since the results show and support the need for further study of the potential benefits of the use of alloplastic TMJ prostheses in the skeletally immature patient population\textsuperscript{20}, whilst the use of distraction osteogenesis also needs to be explored\textsuperscript{2,3,20,38-41}.

**Limitations**

The main limitation of this paper that TMJ replacement with a custom-made alloplastic material in a single immature patient is reported, however additional reports are needed to confirm this successful result.

**Conclusions**

Alloplastic TMJ TJR surgery is being performed successfully and in relatively high numbers by oral and maxillofacial surgeons worldwide with an increasing demand for TMJ TJR surgery in the coming years. In this report, a novel custom-made system, which was produced utilizing CADCAM planning and 3D printing, was introduced. According to the results of this clinical case study, replacement of TMJ with custom-made alloplastic material can be considered as a safe and useful option for growing young individuals in selected cases. However, it is not possible to reach a conclusion with a single report, since there is still a lack of reports in literature. Further studies should be conducted with a larger number of patients with long follow-up periods.

**Conflict of Interest**

The authors declare no potential conflicts of interest with respect to the authorship and/or publication of this article.

**Ethics Approval**

Not applicable for a case report.

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**Authors’ Contribution**

F.G., C.M., M.M., G.B., M.D.F., A.B.G., F.R.P.B., A.B., and D.S.R. revised the work critically for intellectual content. Integrity of the work was appropriately investigated and resolved by all authors. All authors contributed and approved equally to the final version of the manuscript.

**Informed Consent**

A written informed consent form for the treatment in compliance with the principles of Declaration of Helsinki was provided from the patient before the surgery for the treatment. Additional informed consent was obtained from the patient and the parents for using data including photos for scientific purposes.

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