



Case Report

# Custom-Made Alloplastic Prosthetic Implant to Treat Temporomandibular Joint Ankylosis in Pediatric Patients: A Case Study

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**Abstract:** Temporomandibular Joint (TMJ) ankylosis represents a fairly common condition which surgeons need to face. According to Rowe, it can be defined as a restricted functional capacity of the jaw with limited movements owing to bony or fibrous adhesions between the condyle and either glenoid fossa, disc or eminence (or both). It can become a disease which impacts patients' daily life, who suffer from limited mandibular excursion with reduced intercisal opening, anterior open bite, inability to swallow, sleep disorders, and speech problems. In children this may also result in abnormal mandibular and facial growth. In this paper, we deal with the case of a child with an important momolateral ankylosis, previously treated surgically with Costochondral Grafting arthroplasty without success and subsequently treated with a custom-made prosthesis.

Keywords: TMJ; surgery; maxillofacial; ankylosis; pediatric



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## 1. Background

TMJ ankylosis is usually investigated by clinical examination and with imaging studies such as CT or RMI [1–5]. TMJ ankylosis has been categorized into four different entities by Sawnhey et al., taking into account the severity of the situation and recommending a specific surgical treatment based on the type of the defect: Type I: Presence of fibroadhesions at the condyle; Type II: Bone fusion with condyle remodeling and an intact medial pole; Type III: Ankylotic mass, mandibular ramus union with the zygomatic arch and medial pole intact; Type IV: Complete ankylotic mass, total union of the mandibular ramus with the zygomatic arch [4,6–9].

Trauma is the leading cause of TMJ ankylosis globally. In pediatric patients this may be due to falls or even peripartum injuries due to the use of forceps or other obstetric devices, provoking an hemarthrosis mechanism that results in ankylosis [10]. Other etiological factors that can be the cause of this pathological condition are, regardless of age, infections, especially those involving the middle ear, and others, such as rheumatoid arthritis, neoplastic processes, congenital anomalies and recurrent temporomandibular joint ankylosis [11–14].

As mentioned before, TMJ ankylosis usually leads to a series of facial deformities due to the impaired mandibular growth processes, such as retrognathia and retrogenia, if bilateral [11,15,16].

If the deformity is asymmetrical and the ankylosis involves just one side, this abnormality of the mandible's growth process commonly progresses because of two factors: the loss of the condylar fibrocartilage and the inactivity of the joint itself. The condylar growth cartilage does not represent the primary growth site of the mandible, but it is certainly

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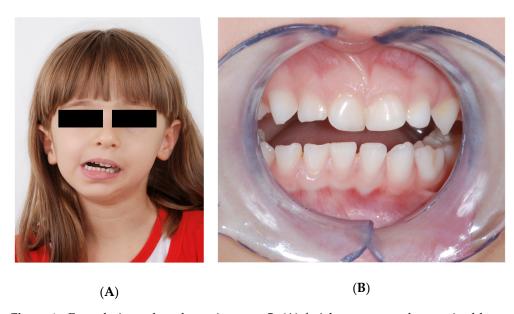
crucial for the correct development of the condylar head, neck, and the ramus to the angle of the mandible. Similarly to other joints, this inevitably provokes an inhibition of condylar linear and cross-sectional growth.

There are several surgical techniques that could be applied to treat TMJ ankylosis in pediatric patients: Costochondral Grafting; Gap arthroplasty or Interposition Gap arthroplasty; Bone distraction; Coronoidectomies; and Total joint replacement, which is the common final stage correction surgery, both with alloplastic custom-made or stock prosthesis [10,14,17,18]. The timing and the ideal sequence of the various interventions is still debated. This is particularly true in subjects with craniofacial deformations, also syndromic, requiring multiple reconstructive procedures where incidence, risk factors, clinical course and long-term outcomes are mostly unknown. The success of these procedures is determined by the maintenance of maximal incisal opening, the prevention of reankylosis and the stimulation of mandible growth [19].

Here we present a case of a pediatric patient who underwent surgery at our clinic. This patient had already undergone previous surgery to remove the ankylotic block and restore the joint functionality, without complete treatment success. It was therefore decided to make her a candidate for a custom-made alloplastic prosthetic reconstruction of the TMJ.

#### 2. Case Presentation

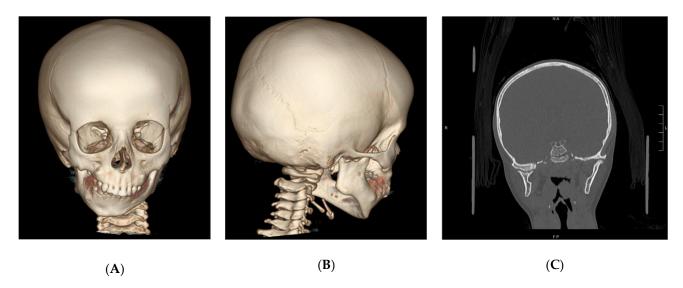
A 5-year-old girl was admitted to the Department of Maxillofacial Surgery at Academic Hospital of Udine, presenting with right TMJ ankylosis. TMJ ankylosis was probably a consequence of a Staphylococcus Aureus sepsis the little girl had suffered when she was just 1 week old. On initial clinical examination, she showed a facial asymmetry characterized by a right deviation of the mandible and the chin, a relatively absent mandibular movement with a maximum mouth opening (MMO; maximum interincisal distance) of less than 10 mm (Figure 1).



**Figure 1.** Frontal view when the patient was 5; **(A)** facial asymmetry characterized by a right deviation; **(B)** Maximum mouth opening was less than 10 mm.

A CT scan performed two years before our first visit showed dysmorphism of the right mandibular condyle with hypoplasia of the condylar neck, dysmorphism of the condylar head and hypertrophy of the coronoid process (Figure 2).

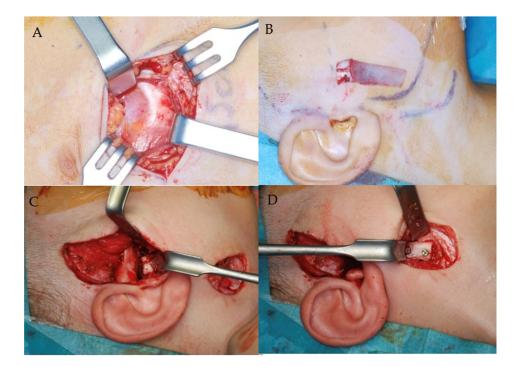
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**Figure 2.** First radiographic evaluation: (**A,B**) Preoperative 3D CT scan, showing the shortness of the condylar neck and the resulting right deviation; (**C**) CT scan coronal section showing the ankylotic bloc.

Due to the patient's age at that time, it was decided to delay the surgery for a couple of years.

Hence surgery was planned, aiming at removing the right ankylotic mass and to perform a contextual reconstruction using a costochondral graft. The right TMJ was approached through a preauricular incision, allowing the excision of the ankylotic block. The costochondral graft was harvested from the fifth rib of the right side and was placed, once shaped, on the condyle area and fixed with two bicortical screws on the right mandible ramus through a retromandibular approach. This was performed carefully to avoid the separation of the cartilaginous part of the graft from the bone. Surgery was performed with no complications (Figure 3).



**Figure 3.** (**A**,**B**) The CCG was harvested from the fifth right rib and then modeled to the exact shape and dimension needed; (**C**,**D**) The CCG is then fixed into place to form the new joint.

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After a first relief of the symptoms and a satisfactory maximum opening of the mouth (MMO), after 6 months the patient began to experience a worsening of the clinical situation which can also be seen in the postoperative radiographic examinations. A recurrence of ankylosis in the right temporomandibular joint was then evaluated, with an MMO of 10 mm, severely limiting normal daily activities and vital functions such as eating, breathing orally and speaking. (Figure 4).



**Figure 4.** (A) MMO of less than 10 mm, (B) evidence of a recurrence of ankylosis.

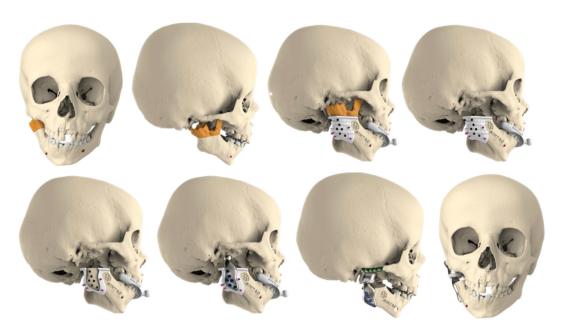
A new surgical plan was then carried out, consisting in the ankylotic block removal, left coronoidectomy and TMJ alloplastic custom-made prosthesis. A new CT scan was performed, and the patient also underwent an intraoral scanning procedure, allowing us to achieve detailed morphology of dental cusps. Moreover, considering the risk of ankylotic bloc removal due to the close relationship with the internal maxillary artery, a CT angiography was performed to evaluate the vascular structures medial to the ankylosis.

DICOM data were processed in bioengineering software (Mimics Innovation Suite, Materialise, Leuven, BE), CT slices were segmented and a mask corresponding to the bone anatomy was yielded, allowing the reconstruction of an entirely digital replica of the patient's skull. Dental cusps derived from intraoral scanning were then merged with those obtained from the CT scan, with substantial improvement of their detail. Then, in a collegial session with expert surgeons, the subcondylar osteotomy was planned bilaterally to balance the contralateral mandibular branch, allowing for the virtual excision of the ankylotic bloc. Moreover, angio-CT data were also processed to achieve the virtual reconstruction of the maxillary artery, whose spatial relationship was assessed in conjunction with the ankylotic bloc removal.

Subsequently, the mandible was put into the correct occlusion with the maxillary arch. Based on the correct occlusion, STL files of the maxilla and the mandible were sent to the manufacturer (Sintac S.r.L, Trento, Italy) for prostheses design. To assist the correct phase of mandible repositioning after ankylosis removal, a surgical splint was also designed.

The TMJ prostheses were designed and validated by the surgical team in a collegiate web session, then the implants were received and sterilized using hydrogen peroxide (Figures 5 and 6).

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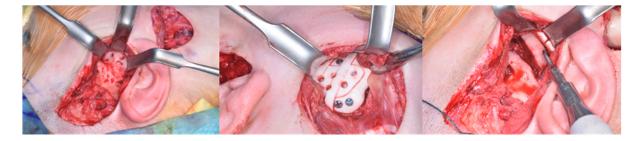
**Figure 5.** Virtual surgical planning. The mandible was isolated to define the resection plane and subsequently design specific cutting guides and the custom-made prosthesis.



**Figure 6.** Fitting test of the custom-made prosthesis on the 3D printed model of the patient's cranium.

The TMJ was exposed by a pre-auricular approach extended to the temporal region, and a sub-mandibular one. A blunt dissection through the submandibular approach was performed until the mandible ramus was exposed. Dissection was carried out as well from the preauricular access, following the cartilage of the external auditory canal until it joined the subfascial pathway. Detachment of the zygomatic arch and evidence of the right TMJ were overturned by severe ankylosis in outcomes of previous CCG surgery (Figure 7).

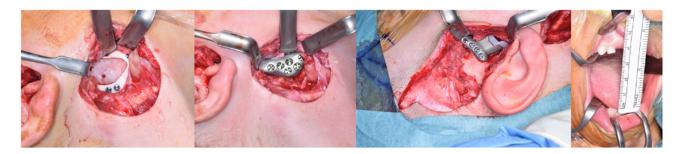
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**Figure 7.** The ankylotic block was identified and excised using the surgical guide and a piezo-surgey device.

The custom-made surgical guide was positioned through the sub-mandibular access and then fixed to the angle and the branch of the mandible with screws. Therefore, we proceeded with a piezoelectric osteotomy [20,21] of the mandibular branch right below the sigmoid incision and a right coronoidectomy.

Simultaneously, through an intraoral vestibular incision in the molar region followed by a subperiosteal skeletonization, a left coronoidectomy was also performed. At this point, the glenoid fossa prosthesis was positioned and fixed with screws, and the condylar prosthesis was covered by fat collected from the periumbilical region to avoid a skin decompression. We proceeded by checking the intraoperatory MMO, resulting a satisfying 35 mm (Figure 8).



**Figure 8.** The custom-made prosthesis was then positioned and the MMO was checked.

After the TMJ replacement procedure was completed, the TMJ accesses were preliminarily sutured and draped to isolate them from the operative field. Then, the mouth was opened and the occlusion was evaluated. In addition, an intraoperative occlusal scan was performed to immediately produce an accurate assessment of the occlusion and a map of precontacts.

Broad spectrum antibiotics and pain meds were administered for 1 week. The patient was placed on a soft diet for 5 days and encouraged to resume a normal diet as soon possible. Physiotherapy was started on the first day post operation, and the patient was encouraged to follow the exercise program at the quickest pace possible. This required following for at least 1 year, perhaps more, availing also of the regular use of *Therabite jaw motion rehab system*, which may contribute to the maintenance of a proper postoperative MMO. A CT scan was performed about 1 week postoperatively, in order to check the position and the alignment of the prosthetic devices to the bony stumps, as well as the relation between the fixation screws and the inferior alveolar nerve. Six months after surgery, at the clinical examination, the patient presented a 25 mm MMO, augmentable up to 30 mm if forced (Figure 9). An annual radiographic check was carried out, useful to intercept any possible complications which may deserve further investigation, but this was not the case. A teleconsultation was performed 14 months after surgery, due to the COVID-19 outbreak [22–25], and the patient showed no significant clinical changes.

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Figure 9. MMO of 25 mm 6 months after TMJ prosthetic replacement.

#### 3. Discussion

This case report aims to report, analyze and introduce the role of personalized alloplastic total TMJ replacement in the sequential management of TMJ ankylosis in early childhood, in this case caused by perinatal Staphylococcus Aureus sepsis. Pediatric TMJ ankylosis should be best viewed as a separate entity due to the lack of consensus on its surgical treatment, the importance of postoperative physiotherapy, and the growth potential of the jaw. Few other features must lead us to think about differences in the management of TMJ ankylosis in children and adults [24]. First of all, as mentioned before, pediatric patients, unlike adults, are growing individuals, which also represents a further element of difficulty in treatment [25,26]. Second, ankylosis and subsequent dentofacial deformities are usually less severe in adults, making their contextual correction easier than in pediatric patients. Third, the appearance of the face and the limitations of opening the mouth could affect the psychological state and the ability to establish social relationships of children.

The timing of the surgery represents an essential topic, taking also in account the necessity of postoperative cooperation of the patient. Kaban et al. suggested that children of 3 years of age or older represent suitable candidates for ankylosis removal, while Yew recommended a minimum surgery eligibility age of 7 years old, considering the injury risks and the compliance of the patient [26]. In our department, pediatric patients affected by TMJ ankylosis are always advised to undergo surgery as soon as possible, usually at 5–7 years old, depending on the compliance of patient and of the family. There is no reason to wait until the end of the growth process to plan the surgery. Delayed treatment may simply worsen psychosocial and physical problems deriving from a moderate to severe facial deformity, and a consequential decrease of life quality.

CCG is generally considered as the first choice for the treatment of this disease in the pediatrics, as it was for our patient [18]. This is because of the ease of harvesting and adapting the graft, the low morbidity of the donor site, the biological and anatomical similarity with the mandibular condyle and its growth potential. Thanks to this development capacity, the CCG theoretically should permit the unaffected side to maintain the same growth pattern, maintaining therefore mandibular symmetry throughout the whole patient's growth process [27–29]. However, the graft growth process could be uncontrolled and unpredictable, ultimately ending in facial asymmetry causing lifelong functional and aesthetic defects. Long-term reports of mandibular growth in children with reconstructed TMJs using CCG shows excessive growth on the treated side occurring in 54% of the 72 cases evaluated, and only 38% of the cases presented uniform growth on both sides, and a relapse of the ankylosis on the recipient site is far from rare [30].

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Graft resorption is another complication that could occur post-operatively, due to poor vascularization of the recipient site secondary to the fibrotic process causing the disease, which may increase the risk of fracture or displacement of the graft itself. Donor site morbidity risks are minimum, but include pneumothorax, infection and chest deformity [31].

Early mobilization and aggressive physiotherapy should be initiated in the immediate post-operative period for patients undergoing GCC surgery, immediately after the release of the intermaxillary fixation (IMF) [32]. In the case of our patient, although proper healing was expected, clinical and radiographic evidences of an ankylosis relapse on the recipient site were highlighted 6 months postoperatively, probably caused by a lack of compliance on proper physiotherapy. At this point the surgical treatment options were few, and the possibility of performing an alloplastic custom-made total joint replacement began to be more consistent.

There is no standardized protocol for the management of TMJ ankylosis, and even less so in pediatric patients. Thanks to scientific advances in materials since 1990s, alloplastic temporomandibular joint replacement has begun to represent a viable option for multiple-operated patients with distorted TMJ anatomy or severe anatomical discrepancies involving the TMJ and recurrent ankylosis. The application of alloplastic TMJ prosthesis has been extensively reported in the adults, but their use in skeletally immature patients is controversial. The main concern regards the interference with the physiological mandible growth process, resulting in an impaired functionality and abnormal shape due to the presence of screws tightened on the mandibular angle which oppose resistance on the downward growth of the mandible ramus [33].

However, reestablishing a correct functionality of the stomatograthic system is essential to drive the correct growth of the maxilla and prevent abnormalities in the occlusal plane. It is well acknowledged that hypoplasia of the affected mandibular side might also result in an asymmetrical growth pattern of the corresponding half of the maxilla. Moreover, the affected side also negatively conditions the development of musculature, resulting in the atrophy of the masseter and the internal pterygoid muscles, which are less stretchable and more fibrotic. Therefore, in the absence of a correct restoration of symmetry and function, the subsequent surgery would bear a substantially higher risk of failure owing to the tendency of muscles to relapse and midface asymmetry which would likely require orthograthic surgery as well, not to mention the psychosocial and emotional implications that the missed restoration of facial harmony and function would have on the child patient [34].

In order to minimize the risk of re-ankylosis, an ipsilateral coronoidectomy was planned at the same time as the alloplastic TMJ replacement, as described by Gerbino et al. [35].

Prostheses are designed based on the final occlusion of the patient, which is virtually determined, therefore virtual surgical planning allows the tailoring of the shape and position of implants on the correct occlusal relationship. Surgical guides are a useful tool to replicate the desired position of prostheses in the real patient, thanks to their strict adherence to the patient's anatomy and their ability to provide predrilled holes and an "installation socket", which further enhances accuracy of placement [35].

Naturally, the major concern regards the need to ensure the patient is a viable candidate for at least one further surgery, considering both the physiological growth of the patient and the potential lifespan of these devices. Cascone et al. suggested three potential solutions for the future surgical revision: (1) Replace the whole prosthesis, or just one of its components, with one of adequate dimensions; (2) A ramus orthognathic procedure, considering that the ramus component of the TMJ total joint replacement device, especially those which are patient-fitted, are fixed posterior to the mandibular foramen; (3) Osteodistraction using the ramus component as one of the legs of the distraction device [36,37].

Adequate physiotherapy and regular follow-up play a leading role in patient follow-up and allow for the detection of any complications related to the first intervention. Furthermore, a careful follow-up of the patient allows the establishment of the correct timing for the second surgery, and the evaluation of when the patient is ready.

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#### 4. Conclusions

The use of a tailored alloplastic TMJ prosthesis for the management of TMJ ankylosis in pediatric patients is controversial, due to a lack of knowledge of potential future growth and function of the jaw. On the other hand, the need for further future surgeries is certain. However, this case represents how this procedure can be useful in selected cases, especially when only a few surgical options are available. Personalized prostheses and virtual surgical planning could also be useful in providing the best functional and aesthetic result to the patient.

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**Informed Consent Statement:** Informed consent was obtained from all subjects involved in the study. Written informed consent has been obtained from the patient to publish this paper.

Data Availability Statement: Not applicable.

Conflicts of Interest: The authors declare no conflict of interest.

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