Review Article

Alloplastic total joint reconstruction for severe temporomandibular joint ankylosis in the skeletally immature patient: a narrative review

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Background and Objective: Temporomandibular joint (TMJ) ankylosis has a severe impact on function and growth. Several treatments have been proposed with limited success and significant morbidity. In the growing patient, alloplastic TMJ reconstruction (TMJR) is usually reserved as the last resource. The purpose of this article was to perform a narrative review to access the feasibility of treating ankylosis in the growing patients with alloplastic implants.

Methods: A comprehensive electronic search was conducted on MEDLINE/PubMed, up to Jun 15, 2022. Medical subject heading terms (MeSH) and free-text words were used in the electronic database searches to find articles that reported: Temporomandibular Joint; OR Temporomandibular AND Joint AND Ankylosis; OR Ankylosis AND Joint Prosthesis; OR Joint AND Prosthesis; OR Alloplastic. The primary outcome was to find how many cases have been reported involving the treatment of ankylosis in the skeletally immature patient with alloplastic TMJR. Any study that reported the use of these devices to treat ankylosis in patients at age 16 or younger was included.

Key Content and Findings: Two hundred and eighty-eight potential studies were identified for titles and abstracts, and 48 studies were included for full-text reading. Fifteen articles were found with at least 1 report of skeletally immature patients with ankylosis treated with alloplastic TMJR.

Conclusions: Alloplastic TMJR is a good treatment option in growing patients with ankylois that have failed conventional treatment such as gap arthroplasty (GA) and grafts, but it may also be considered as a first line treatment.

Keywords: Alloplastic joint; temporomandibular joint reconstruction (TMJR); ankylosis; growing patient

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Introduction

Alloplastic temporomandibular joint reconstruction (TMJR) surgery has been traditionally reserved for adult patients and usually as a last resource after previous unsuccessful treatments. Surgeons have traditionally regarded open procedures of the TMJ in the growing skeleton with appropriate caution and healthy skepticism especially as it relates to growth or mandibular function. The major indication for operative treatment of the TMJ in adolescent/pre-pubescent patients is pathology, in

particular ankylosis (1-3). Compliance with post-operative rehabilitation is challenging for both the child and parents, and is often abandoned early in the convalescence by the family. This more often than not compromises the range of motion obtained at surgery and can result in re-ankylosis of the affected mandibular condyles, necessitating another operation. Multiple surgery then carries the risk of worsened scarring and possible growth injury resulting in a spiraling decline in mandibular function for the child and concomitant decrease in mastication, skeletal deformity with compensatory deformity of the maxilla, and rampant dental disease leading to premature tooth loss.

The etiologies of TMJ ankylosis were reviewed extensively in 1964 by Topazian (4), and have, for the most part, remained unchanged. Treatment of TMJ ankylosis in adults and children has undergone an evolution, and a subsequent improvement in outcomes. The treatment of pediatric patients includes the following: excision of the ankylosis with a bone gap, growth center transplants, distraction osteogenesis (DO), and recently alloplastic total joint replacement. It should also be noted that early DO of the mandibular ramus can potentially lead to TMJ ankylosis. This is most likely due to poorly planned vectors in the ramus that result when distraction appliances push the small superior ramus component and condyle toward the zygomatic arch and glenoid fossa. This may lead to ankylosis of the condyle, glenoid fossa, and zygomatic arch (5,6).

Perrott and Kaban in 1994 (7) and Katsnelson in 2012 (8) have significantly contributed to the understanding and treatment of TMJ ankylosis. Their reports focused on gap arthroplasty (GA), and found it to be superior to resection followed by condyle-ramus unit reconstruction with a costochondral graft (CCG) (9-11) or sternoclavicular graft (12). Nevertheless, one of the main difficulties in this treatment is that the management of TMJ ankylosis in children is significantly impacted by the etiology of the problem. Additional factors also play a crucial role, including the age of the patient, the involvement of one or both condyles, the magnitude of ankylosis, and the rehabilitation potential of the patient.

Previous studies continue to produce different results, and many treatment modalities have been used with a number of different outcomes with various successes and failures. Sahoo *et al.* (13) reported results treating ankylosis with DO, CCG, and placement of a muscle flap with fat. He reported that while all procedures yielded similar results, he preferred CCG and distraction for the treatment of ankylosis in children. He attributed most failures to a lack of compliance with rehabilitation protocols (14). This was in clear contrast to similar studies performed by Perrott and Katsnelson and, who attributed treatment outcomes to the method of reconstruction (7,8).

Studies done in the past have helped to elucidate the various surgical techniques for treating TMJ ankylosis that take advantage of the growing skeleton. In 1909, Bardenheuer first described his technique of replacing a mandibular condyle with a patient's 4th metatarsal. Nearly 70 years later, Matukas *et al.* (15) described the use of an iliac crest, which was fixed to the zygomatic portion of the temporal bone, followed by placement of the cartilaginous portion facing the stump of the mandibular ramus (15). This procedure provided appropriate mandibular opening for 18 months in a 5-year-old with TMJ ankylosis. Nearly a year later, Ware and Brown (16), described the results of 10 patients, each of whom had undergone growth center transplantation to replace mandibular condyles either due to trauma or degenerative conditions. One such implant was the fibular implant; and although there was a lack of uniformity, there was significant unpredictable growth of the transplants.

More recently, however, some studies have focused on comparing several of these grafts, namely the sternoclavicular graft and the CCG. As first described by Gillies in 1920 (17), the CCG became a popular choice for autogenous transplantation in children with defective mandibular condyles. CCGs are commonly used based on the premise of the biological and anatomical similarity to the condyle. They also have the characteristic of both primary and secondary growth potential in a similar fashion to the human condyle. However, it became apparent that successful continued growth and harmonious function did not occur as often as surgeons had hoped. Reasons for the lack of universal success most likely hinged on the differing growth characteristics between the two joints. Additional disadvantages include fracture of the cartilage, poor bone quality, unpredictable growth, pneumothorax, and the use of a second donor site.

It wasn't until 1986, when Ellis and Carlson postulated that the sternoclavicular graft would be more suitable in TMJR due to the similar developmental and structural origins (18). They stated that the sternoclavicular graft resembled the mandibular condyle cartilaginous structure that is separated by a mesenchymal layer. Nevertheless, this donor site option didn't become very popular (19).

Lastly, many practioners are confronted with poorly

planned and executed mandibular distractions done at a very young age, often repeatedly, which invariably leads to severe recalcitrant ankylosis. In addition, the repeated use of mandibular distraction at a young age causes both significant periosteal/muscle scarring and deformity of the condyle-glenoid fossa unit, which makes autogenous reconstruction all the more difficult. This unpredictability, coupled with the difficulty in reconstruction, has led surgeons to perform alloplastic joint reconstruction in the growing facial skeleton for complicated TMJ ankylosis. The purpose of this article was to perform a literature review to access the feasibility of treating ankylosis in the growing patients with alloplastic implants. We present this article in accordance with the Narrative Review reporting checklist (available at https://fomm.amegroups.com/article/view/10.21037/fomm-22-10/rc).

Methods

The Patient, Intervention, Comparison, Outcome, and Study (PICOS) strategy was not applicable to this review, due of the novelty of this procedure. The primary outcome was to find how many cases have been reported involving the treatment of ankylosis in the skeletally immature patient with alloplastic TMJR. Any study that reported the use of these devices to treat ankylosis in patients at age 16 years old or younger was included.

A comprehensive electronic search was conducted on MEDLINE/PubMed, up to Jun 15, 2022. Medical subject heading terms (MeSH) and free-text words were used in the electronic database searches with the following strategy: (((Temporomandibular Joint[MeSH Terms]) OR ((Temporomandibular) AND (Joint))) AND ((Ankylosis[MeSH Terms]) OR (Ankylos*))) AND (((Joint Prosthesis[MeSH Terms]) OR ((Joint) AND (Prosthes*))) OR (Alloplast*)). A single reviewer FGR screened the retrieved studies for inclusion, based on the titles and abstracts. Relevant research articles were assessed in full for eligibility.

Results

Initially, 288 potential studies were identified. The titles and abstracts were then screened, and 240 studies did not meet the eligibility criteria and were excluded, resulting in 48 studies included for full-text reading. After full-text reading 15 articles were found with at least 1 report of skeletally immature patients with ankylosis treated with alloplastic TMJR (1,2,20-32).

Discussion

The study of TMJ ankylosis at Parkland Hospital can be traced all the way back to 1958 and 1960, when Dr. Walker evaluated both arthroplasty and growth pattern in TMJ ankylosis (33,34). It was his belief that in order to understand the adaptability of the condyles, one should begin with the understanding of how condylar fractures heal. The first paper discussed the use of arthroplasty on the ankylotic joint, while the second paper focused on growth patterns in skeletally immature monkeys with TMJ ankylosis. These landmark papers provided a unique understanding of the adaptability of the mandibular condyles. The complications that arose were often related to the poor quality of rehabilitation (hypomobility, pain, occlusion, and arthritis). In our experience, and many others, physical therapy is absolutely needed for good long-term results.

Back in 1979, Kiehn *et al.* (20) published their attempt to adapt an orthopedic technique (Charnley technique) to the TMJ. This involved 2 preformed Vitallium prosthesis that were adjusted to the base of the skull and the ascending ramus, and fixated to the bone with polymethylmethacrylate (PMMA). The authors reported 27 cases operated, including 7 children. They observed 6 children for 2 years and 1 child for 4 years, and found good results, except that 1 child had the implant eroded through skin. The authors recognized problems during mandibular development and suggested additional surgery with the placement of iliac bone graft in the horizontal ramus, in an attempt to maintain the normal gonial-gnathion distance.

In 1990, Westermark *et al.* (21) reported the case of an 8-year-old girl treated with resection of ankylotic block and insertion of a Delrin device. They were based on promising orthopedic results and an animal study that demonstrated bone apposition around the prosthetic components (35). The TMJR was performed at the same time of ankylosis resection. The new glenoid fossa was lined with lyophilized dura that was sutured to the bone, while the Derlin condyles, fixed in a titanium mesh, were then installed after final occlusion was obtained. The patient was followed for 2 years and demonstrated a forward and downward growth of the maxilla, posterior rotation of the mandible, and open bite. Even though they were expecting bone apposition at the TMJ level to maintain normal mandibular growth, patient ended up with mandibular retrognathism that

required further surgical procedure.

On the following 30 years, there have been sparse reports of surgeons treating TMJ ankylosis in growing patients with alloplastic TMJR. Most of the times this was performed after conventional treatment had failed, and with limited follow-up. Kummoona (22) was probably an exception, and was able to follow up 3 patients that received a 2 parts Chrome-Cobalt prosthesis (fossa + mandibular parts) after resection of TMJ ankylosis. Two patients were operated at 8 years of age, and were followed for 10 years, while 1 patient was operated at 7 years of age and was followed for 15 years. Mouth opening improved significantly in all 3 patients from less than 10 mm to greater than 34 mm, but unfortunately, no comments were made on mandibular development. In 2010, Westermark (23) reported 2 cases of bilateral TMJR with bilateral stock joints in 14 years old males. One of these patients was followed up for 7 years and changed from 13 mm pre-op maximal incisal opening (MIO) to 15 mm post-op, while the other patient changed from 0 mm pre-op MIO to 37 mm after 3 years. No comments were made regarding facial development.

Wolford *et al.* (24) reported no complications in the pediatric population with TMJ ankylosis that was treated with alloplastic TMJR. They consider that females older than 15 years old, and males older than 17 years old can be treated as adults. The author was reached by email in June 2022 and was able to give an update of the 2 patients that received TMJR at 12 years of age. They both required surgery around 18 years old. One patient underwent sagittal split osteotomy on the non-affected side, and detachment of the rams portion of the prosthesis to advance the mandible with reattachment of the mandibular component onto the mandible, in conjunction to maxillary osteotomies for counterclockwise rotation advancement of the maxilla-mandibular complex and correction of the facial asymmetry. The other patient was originally treated with bilateral TMJR, and around 18 years old presented growth vector in a downward and backward direction. Because this patient had received a more standard design configuration prostheses (compared to the first one), Dr. Wolford was able to do bilateral intraoral mandibular sagittal split osteotomies with the prostheses remaining attached to the proximal segment, and maxillary osteotomies for counterclockwise rotation of the maxilla-mandibular complex, with a great result.

Cho *et al.* (25) reported the case of a patient submitted to CCG at 12 years old that failed, GA at 13 years old, TMJR at 15 years old with a stock condyle screwed into a reconstruction plate, and finally ramus reconstruction at 17 years old with iliac bone graft to compensate additional growth of the mandible. Lastly, Sinn *et al.* (26) reported 5 cases of growing patients treated with alloplastic TMJR with a follow-up that varied from 46 to 121 months. All patients improved MIO, and 1 infection was observed which required further surgery.

After evaluating the success of 13 CCG, performed in growing patients to reconstruct the TMJ for multiple etiologies, Ross (36) found a higher success rate in young patients. From ages 3 to 9 years success was 80%, whereas the success was 50% for patients older than 14 years of age. The author's definition of success was chin deviation less than 3 mm, and he reported 46% success, 39% excessive bone growth, and 15% showed deficient growth on the graft. Chen *et al.* (5) found less recurrence with CCG when compared to DO. Of the 23 type III ankyloses cases, 19 were reconstructed with CCG and 14 with DO. Only 1 case (5.3%) relapse on the CCG group, while 4 (28.6%) recurrences occurred in the DO group. Complications associated to the donor site including pain, pleura perforation, pneumothorax, and rib deformity have also been described (37).

Greatest concern with CCG is the unpredictable growth and the reankylosis possibility (38,39). After evaluating 21 CCG performed in 12 pediatric patients with juvenile chronic arthritis (JCA), Svensson and Adell (3) found that 7 (58%) required late orthognathic surgery to correct mandibular prognathism or asymmetry. Furthermore, 3 unilateral cases that did not require further orthognathic surgery, were under corrected at the time of CCG procedure.

Some authors have also demonstrated overgrowth of the CCG in the lateral direction at the level of the condylar head (7,40). Ko *et al.* (40) also found an upright growth pattern with the CCG, meaning that even with similar amounts of increase in mandibular length and ramus height on both grafted and normal sides, an upright and forward position of the grafted condyles would move the chin toward the non-affected side. The authors (40) stress the importance of long-term follow-up. They followed 10 consecutive patients treated for TMJ ankylosis with CCG. The 3 patients that were followed until adolescence growth spur demonstrated overgrowth and required additional surgery.

Keyser *et al.* (1) reviewed 14 cases from TMJ Concepts records of patients between 7 and 17 years that had 24 TMJR performed. Two complications were observed

among the 24 implants: biofilm and heterotopic bone formation in different individuals. The complications were addressed by removal plus replacement of implant, and endoscopic removal of heterotopic bone, respectively. The authors concluded that alloplastic TMJR in growing individuals may have a role in specific situations such as high inflammatory arthritis unresponsive to other modalities of treatment; recurrent fibrosis and/or bony ankylosis unresponsive to other modalities of treatment; failed tissue grafts; and loss of vertical mandibular height and/or occlusal relationship.

It is unclear if the limited growth is due to the alloplastic joint itself or due to severe injuries incurred on soft tissues and bone. Nevertheless, our initial results demonstrate the simplicity of the post-operative rehabilitation and maintenance of improved MIO for these children. In the cases where remaining growth lead to asymmetry, potential solutions are: replace the implant (fossa + mandibular components or just mandibular component); osteotomy on the bone anterior to the implant.

In his landmark 4-part publication, Moss described the unique characteristics that allow for the proper growth and development of the facial skeleton (41-44). The developmental origin of all cranial skeletal elements (e.g., skeletal units), coupled with changes in size, shape (e.g., form), and location are a result of the responses to the function of their surrounding structures, both at the cellular and histologic levels. Therefore, maintaining these positions are also—without exception—secondary to compensatory mechanically obligatory responses toward the demands of their related cephalic non-skeletal cells, tissues, organs and operational volumes (e.g., the functional matrices). Specifically, the function of soft tissues on bone is critical to growth and form of bone, all of which could potentially negatively be affected by ankylosis in the growing child.

Performing a traditional systematic review using the PICOS statement for reporting guidelines is quite challenging if not impossible due to the novelty of the technique and the limited amount of literature available. On top of that, the articles that have been published are mainly reports and descriptive, with limited number of patients that would allow more sophisticates analysis.

The current parameters of care for TMJR in growing patients include techniques described over 40 years ago, that have a less than ideal success rate to provide adequate joint function and facial balance. Complications such as reankylosis, restricted growth, as well as excessive growth, seems to be unpredictable with traditional techniques, often requiring additional surgery. Because it is such a rare situation with multiple etiologies, affecting different age groups, and seemingly very operator sensitive in terms of success rate, it is very hard, if not impossible to perform a controlled trial comparing CCGs, osteogenic distraction, free fibula flap, and alloplastic joints to reconstruct the TMJ in growing patients. Expert opinion of highly experienced surgeons seems to be the best evidence at this point. Most authors seem to agree that alloplastic TMJR may be used as a rescue procedure in growing patients after failure of allograft (2,27).

Conclusions

There is significant controversy surrounding the use of alloplastic total joint reconstruction in the growing patient as the first treatment line. Most surgeons prefer to reserve this technique as a backup plan for refractory ankylosis or multiple failed surgeries. Although there is concern about its place in children with significant growth potential, it should be noted that children with TMJ ankylosis already lack the mandibular growth potential seen in children without this condition.

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Footnote

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