

**FIGURE 4.** (A) Six-month follow-up showing stability in the facial midline and contouring symmetry. (B) Computed tomography showing internal rigid fixation with skeletal symmetry and the TMJ prosthesis with no complications. (C) Orthopantomography showing the position of the TMJ prosthesis and the osteotomy in the basilar border of the left mandibular angle and body. TMJ indicates temporomandibular joint.

TMJ-R and mandibular osteotomy (lower border of the condylar component) (Fig. 4) obtaining.

The mandibular contouring should be included in the digital workflow when preparing a TMJ-R after low condylectomy and orthognathic surgery; this is a critical point to obtain facial symmetry and facial esthetic after surgical treatment.

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OPEN

# Surgical Treatment of Temporomandibular Joint Ankylosis: our experience with 36 cases

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**Aim:** To evaluate and compare outcomes of patients with temporomandibular joint ankylosis (TMJA) treated by gap arthroplasty, costochondral graft, and total alloplastic joint reconstruction.

**Methodology:** A retrospective cohort study reviewed and analyzed data from patients with TMJA from January 1, 2009 to December 31, 2019, at the Maxillofacial and Oral Surgery Department, University of the Witwatersrand. Patients with TMJA were treated either with gap arthroplasty, costochondral graft, or total alloplastic joint reconstruction. Data collected included age, sex, etiology of ankylosis, sides involved, pre-operation and postoperation mouth opening (MO), treatment type, complications, and revision surgery. Patients were followed up for at least 18 months after the surgical procedure. Comparison of means across the treatment groups was analyzed using paired *t* tests or analysis of variance test. A *P* value of less than 0.05 was considered statistically significant.

**Results:** The study sample comprised of 36 patients [bilateral, *n* = 22; unilateral, *n* = 14 (21 male, 15 female)]. Trauma was the most common etiology (*n* = 27, 75%), followed by chronic infections (*n* = 4, 11.11%) and juvenile arthritis (*n* = 3, 8.3%). A paired *t* test revealed no statistical significance between treatment modality and postoperative MO and complications over 18 months (*P* = 0.5316 and *P* = 0.426, respectively). The mean MO increased from 4 to 28 mm. Reankylosis was the most common complication (*n* = 5).

**Conclusions:** All 3 treatment options yield acceptable outcomes in patients with TMJA. Irrespective of surgical technique, early postoperative exercises, active physiotherapy, and follow-up are imperative for successful rehabilitation and prevention of reankylosis.

**Key Words:** Alloplastic joint reconstruction, costochondral graft, gap arthroplasty, temporomandibular joint ankylosis

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Temporomandibular joint ankylosis (TMJA) is a debilitating mandibular hypomobility disorder characterized by a fusion of the condyle to the fossa by bony or fibrotic tissues. Temporomandibular joint ankylosis may be associated with serious functional and esthetic deformities, including limited mouth opening (MO), impaired mastication, malnutrition, poor oral hygiene, and disturbances of facial and mandibular growth.<sup>1</sup> Mandibular hypomobility and subsequent retrognathia can produce oropharyngeal airspace narrowing, leading to signs and symptoms of obstructive sleep apnea syndrome.<sup>2,3</sup>

Temporomandibular joint ankylosis may be acquired or congenital. Acquired TMJA is most commonly associated with trauma, local/systemic infections, previous TMJ surgery or systemic diseases such as ankylosing spondylitis, rheumatoid arthritis, and psoriasis.<sup>4-11</sup>

The ultimate treatment goal of TMJA is to restore altered joint mechanics, correct the associated dentofacial deformities, facilitate maintenance of good oral hygiene, and prevent reankylosis.<sup>12</sup> Various treatment modalities for TMJA have been described, and these include gap arthroplasty (GA), interpositional gap arthroplasty, and reconstructive arthroplasty (RA) (total joint reconstruction with autogenous grafts or alloplastic materials) and distraction osteogenesis. Interpositional gap arthroplasty involves resection of the osseous mass followed by interpositioning of a biological or alloplastic material in the ensuing gap.<sup>13-21</sup> In RA, the ankylosed mass is resected and the joint is reconstructed by autogenous bone grafts or a total alloplastic joint prostheses.<sup>13,14,20,22</sup>

However, there is no accepted standard surgical protocol in TMJA surgery that has produced consistent results, and the measurement of outcomes is often not clear or standardized.

The purpose of this study was to evaluate and compare outcomes of patients with TMJA treated by 2 surgical modalities: GA and RA. The specific aims of the present study were to compare the preoperative and postoperative MO, and to determine complications associated with each treatment modality.

## STUDY METHODOLOGY

A retrospective cohort study was conducted to review and analyze records of patients with TMJA who were treated at the Department of Maxillofacial and Oral Surgery, Chris Hani Baragwanath Academic Hospital, University of the Witwatersrand (Johannesburg, South Africa) between 2009 and 2019. The study was approved by the University's Human Research Ethics Committee (M230130), and written informed consent was obtained from all patients.

All patients who had at least 18 months of follow-up after surgical management of TMJA were included in the study. The exclusion criteria included all patients who had failed to return for the requisite 18-month follow-up and patients with missing records.

Mouth opening and complications such as reankylosis were the primary outcome variables. Secondary outcome variables included demographic variables, etiology of ankylosis, laterality, treatment modality, preoperative MO, and revision surgery. Treatment consisted of resection of ankylosed mass (GA alone) through the preauricular approach or GA followed by reconstruction with costochondral graft (CCG) or stock total alloplastic joint reconstruction (TAJR). In GA alone, a 1 to 1.5 cm gap was created after resection of the ankylosed mass and left without interpositional material.

For RA, a combination of the preauricular with temporal extension and Risdon approaches were used to expose the TMJ and the mandibular ramus, respectively. After GA, the MO

measured up to 35 to 40 mm in all patients. If there was a limitation in MO of less than 35 mm, ipsilateral followed by contralateral coronoidectomy, then pterygomasseteric sling release was also performed until the patient's MO of 35 mm was reached. The graft was harvested through a submammary incision from the fifth/sixth ribs for CCG. The thickness of the cartilaginous component ranged from 3 to 5 mm. Rigid internal fixation of the CCG was provided by 2 to 4 bicortical screws secured on the mandibular ramus. Postoperative intermaxillary fixation was established for 6 weeks to stabilize the graft and promote optimal healing.

For TAJR, each patient underwent a standard protocol regarding the work-up for reconstruction with stock joints that involved both the condylar and fossa components. The surgical procedures also involved complete ankylosis removal and coronoidectomies. Following TAJR, the mandible was then manipulated to ensure proper mating between the fossa and the condylar prostheses and to confirm that there are no mechanical obstructions or anterior dislocations. Mouth opening exercises were commenced on day 2 postoperatively.

Next, patients were referred for intensive physiotherapy after the removal of the fixation. They were instructed to practice MO exercises with sticks or a TheraBite Jaw Rehabilitation System (Atos Medical, Horby, Sweden). Physiotherapy was started within 24 hours in GA and TMJ alloplastic reconstruction groups and soon after, a period of intermaxillary fixation in CCG group.

The primary outcome variables were maximum MO and complications during follow-up.

## Statistical Analysis

Data was captured into a Microsoft Excel spreadsheet (Microsoft Office 365; Microsoft, Redmond, WA) and cleaned to allow for analysis using Stata IC/14 software.

Measured variables (sex, age, laterality, preoperative, and postoperative MO) were presented as mean (standard deviation).

The patients were categorized into different groups based on the sex, laterality, and treatment modality chosen. Preoperative and postoperative data were analyzed and compared. The comparison of means across the groups was analyzed using paired *t* tests or the analysis of variance test. A *P* value of less than 0.05 was considered statistically significant.

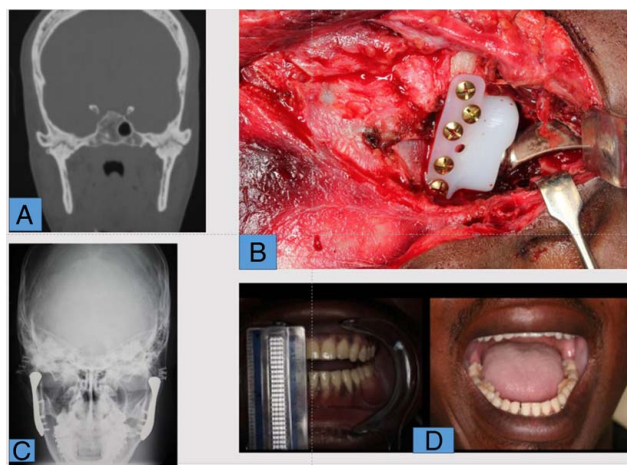
## RESULTS

A total of 51 patients with TMJA were enrolled in the study, of which met the inclusion criteria (15 females and 21 males). The mean age was 24 years (range: 3–70 y), and those younger than 20 years of age were the most frequent (*n* = 18).

Thirty-nine percent (*n* = 14) of patients had unilateral ankylosis, while 61% (*n* = 22) had bilateral ankylosis (BA). Overall, trauma was the most common etiology (*n* = 27), followed by chronic infections (*n* = 4), juvenile arthritis (*n* = 3), burns (*n* = 1), and 1 unknown. Nineteen (70%) of the 27 patients who reported trauma as the causative factor presented with BA.

Preoperative MO ranged from 0 to 10 mm (mean = 5 mm). There was no statistical significance in preoperative MO among the different age groups (*P* value = 0.1162). However, there was statistical significance in preoperative MO between males (*P* value = 0.0084) and in unilateral cases when compared with bilateral cases (*P* value = 0.0048).

Total alloplastic joint reconstruction was the most common treatment modality, followed by CCG (Table 1, Supplemental, Supplemental Digital Content 1, <http://links.lww.com/SCS/G134>). Figure 1A-D shows a coronal computed tomography



**FIGURE 1.** Images showing (A) coronal CT scan in a patient with bilateral temporomandibular joint ankylosis, (B) intraoperative photograph showing alloplastic joint with the condylar head in the fossa, (C) postoperative x-ray posteroanterior (PA) mandible showing bilateral joints, and (D) preoperative and postoperative mouth opening after 1 year.

scan, TAJR in situ, postoperative posteroanterior mandible x-ray, and preoperative and postoperative MO, respectively, of a patient with BA. Figure 2 shows a 1-year postoperative panoramic radiograph showing bilateral joints.

Postoperative MO at 18 months ranged from 15 to 40 mm. Age group, laterality, and treatment modality were not significantly associated with this outcome ( $P=0.5791$ ,  $0.8204$ ,  $0.5316$ , respectively) (Tables 2, Supplemental Digital Content 2, <http://links.lww.com/SCS/G135> and 3, Supplemental, Supplemental Digital Content 3, <http://links.lww.com/SCS/G136>). However, the study showed that males had significantly better MO outcomes compared with females ( $P$  value =  $0.0051$ ).

The complication difference between the treatment groups was not statistically significant ( $P=0.426$ ). The most common postoperative complications were reankylosis ( $n=5$ ) and heterotopic bone formation ( $n=3$ ) (Table 4, Supplemental, Supplemental Digital Content 4, <http://links.lww.com/SCS/G137>). Three patients had weakness of the temporal branch of the facial nerve, from which 2 had complete resolution.

Nine of the 13 patients who had postoperative complications subsequently had revision surgeries. Four of the 5 cases with postoperative reankylosis were managed with excision of the ankylotic mass, revision of GA, and TAJR, while 1 patient had unilateral GA only.

Two of the 3 patients who had heterotopic bone formation (HBF) were managed with the removal of heterotopic bone,



**FIGURE 2.** A 1-year postoperative panoramic radiograph showing bilateral alloplastic joints following release of temporomandibular joint ankylosis.

stretching of the jaw and re-implantation of the prostheses, while 1 was managed with removal of the prosthesis on 1 side and replacement with a new alloplastic prosthesis.

One patient who was treated with bilateral CCGs later presented with severe malocclusion in addition to asymmetry and unilateral reankylosis. He was treated with a combination of excision of bilateral ankylotic mass, TAJR, and orthognathic surgery to correct both asymmetry and malocclusion.

## DISCUSSION

We designed this study to evaluate and compare the outcomes of patients with TMJA treated by GA, CCG, and TAJR. The specific aims of the present study were to compare the preoperative and postoperative MO, and to determine complications associated with each treatment modality.

After a follow-up of 18 months, no significant differences in MO and complications were observed between GA, CCG, and TAJR. Males, however, had significantly better MO outcomes compared with females.

Notably, trauma was the most common etiological factor, in concordance with previous studies, confirming maxillofacial trauma as the main cause of TMJA.<sup>18,19</sup> Early diagnosis and management of trauma, particularly in children, is of paramount importance in the prevention of TMJA. However, lack of resources and low socioeconomic status in developing countries often precludes this.<sup>20</sup>

Though not significant, both GA and TAJR yielded better MO than CCG. Roychoudhury et al<sup>21</sup> also reported that improvement in MO was more in the GA group than CCG after a 12-month follow-up. They explained that the post-GA shift of the fulcrum of opening to either mid-ramus or last standing tooth (especially in bilateral cases) aids in large MO. In CCG, the fulcrum remains in the condylar area due to ramus-condyle unit reconstruction and this may hinder larger movements; however, the MO achieved is adequate and within normal limits. Westernmark<sup>22</sup> and Aagaard and Thygesen<sup>23</sup> reported that the long period of ankylosis (from childhood for many of these patients) affects the stretching capabilities of all surrounding tissue components. They concluded that it is almost impossible, under these conditions, to restore normal opening of the jaws and suggested that a MO greater than 25 mm and maintained permanently could be considered a success. Stripping of the pterygomasseteric sling combined with the coronoidectomy greatly contributes to improved MO. Khalifa<sup>24</sup> reported that monitoring MO for up to 1 year postoperatively, at least, is very important to detect early signs of reankylosis that are not shown in radiographs. We concur that a decrease in MO does not always imply recurrence but may manifest poor compliance and failure to adhere to the prescribed physiotherapy protocol.<sup>25</sup>

Of all surgical techniques for TMJA, GA is reported to have the highest risk of recurrences, with reankylosis occurring even after following very wide GA.<sup>24-26</sup> Gap arthroplasty alone in bilateral cases may result in anterior open bites leading to the development or worsening of pre-existing mild to moderate obstructive sleep apnea.<sup>27</sup> Because of its inherent problems, GA alone has largely been abandoned as a definitive treatment modality for TMJA and replaced by a protocol interposing a lining material (preferably autogenous) between resected surfaces.<sup>8</sup> Notwithstanding this, GA was chosen in this study in only unilateral cases because it is a cheap and less complex treatment modality. We also concur with others that regardless of the surgical technique in TMJA, early mobilization, aggressive physiotherapy, and good cooperation from the patient



are widely regarded as important factors in preventing reankylosis.<sup>24</sup> In the present study, no case of reankylosis was observed following GA. However, a longer follow-up beyond 18 months could have yielded a different outcome.

This study's finding appear to indicate that TAJR has gained popularity over CCG when TMJ reconstruction is indicated; and is now considered the gold standard in reconstruction of the irreparably damaged adult TMJ, ankylosis in particular.<sup>4</sup> The major advantage of using TMJR in TMJA is that it allows for immediate reconstruction and function, including the possibility of early aggressive physiotherapy, which is considered essential for rehabilitation and long-term success and an important factor in preventing recurrence.<sup>28</sup> Mean preoperative MO of 14.5 and 11.75 mm and postoperative MO of 35 and 32.9 mm, respectively, were reported by Wolford et al,<sup>5</sup> and Mercuri et al<sup>29</sup> after treating TMJA with TAJR. Their techniques included periarticular fat grafting around the prostheses. The rationale for placing autologous fat grafts around the TMJ prosthesis is to obliterate the dead space surrounding the joint prosthesis, thus preventing the formation and subsequent organization of a blood clot. Creating this physical barrier, the fat graft minimizes the presence of pluripotential cells and prevents formation of extensive fibrosis and heterotopic calcification.<sup>5</sup> HBF leading to reankylosis has been a problem after arthroplasty, autogenous, and alloplastic total TMJ reconstruction, particularly in recurrent cases. We observed HBF requiring removal and re-implantation in 3 patients treated with TAJR. For patients at risk of HBF (such as those requiring revision of failed autogenous/alloplastic TMJR devices, multiply operated joints, and ankylosing spondylitis), there is plausible justification for incorporation of the periarticular autogenous abdominal fat grafting protocol. Other methods used to minimize HBF are low-dose radiations, nonsteroidal anti-inflammatory drugs (indomethacin), and copious irrigation with saline to remove all the bone chips/slurry to prevent the impregnation of bone morphogenetic protein in surrounding tissue.<sup>18</sup>

In the present study, 5 of the 9 patients treated with CCG suffered reankylosis, and 1 had malocclusion resulting from excessive growth. Unpredictable excessive growth and reankylosis of CCG remain the biggest drawback of TMJ reconstruction in the growing patient. Transplantation of an excessive amounts of cartilage is thought to increase the risk of complications in CCG, in particular reankylosis and unpredictable excessive growth.<sup>8,12,30</sup> Dimitroulis<sup>30</sup> also reported that CCGs were more likely to require re-operation either for reankylosis or to correct excessive growth. Following treatment of 95 patients with CCG, Wadde et al<sup>19</sup> reported complications such as reankylosis (6.32%), graft overgrowth (13.70%), insufficient graft growth (22.11%), no graft growth (3.20%), and facial asymmetry (20%). They concluded that factors such as graft cartilage thickness and use of interpositional material or lack thereof could be correlated with these complications. It appears like the ankylotic milieu that the CCG finds itself, aggravated by muscle atrophy, fibrosis of the ligaments, and lack of aggressive physiotherapy, provide a fertile ground for growth abnormalities in CCG cases.

The present study has multiple limitations. First, the type and morphology of ankyloses were not specified. Second, there was no standardized physiotherapy protocol (method and duration). In addition to a diverse age group, we included patients with not only acquired but congenital TMJA and those with systemic diseases affecting the joints, that is, juvenile rheumatoid arthritis, thus creating a non-homogenous group. It was also not possible to determine the duration of ankylosis from the patients' history.

## CONCLUSIONS AND RECOMMENDATIONS

From the present study's findings, revealing no significant difference in postoperative MO and complications between GA, CCG, and TAJR, we conclude that all 3 treatment modalities provide acceptable outcomes in patients with TMJA. Early surgical intervention, radical and sufficient resection of the ankylosed mass, early postoperative exercises, active physiotherapy, and follow-up, are imperative for successful rehabilitation and prevention of reankylosis. Future prospective randomized-controlled studies, involving a larger number of patients, and longer follow-up periods that will include data on the type of ankylosis and standardized physiotherapy protocol, are recommended to further strengthen the findings of this study.

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## The Long-Term Natural History of Fibrous Dysplasia

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**Abstract:** Fibrous dysplasia (FD), a developmental, nonfamilial, benign anomaly of bone development, is characterized by the replacement of normal bone by proliferating fibro-osseous tissue. Marked craniofacial deformities, functional disturbances, and emotional stress are major indications for treatment, and various surgical procedures have been performed; however, excision and regrowth issues have also been reported. While several treatment options are available, no studies have reported the natural history of untreated FD. Here, we report 2 patients, aged 73 and 50 years, respectively, who had not received treatment. Both patients presented to the hospital complaining of noise when moving their heads. Computed tomography scans showed niveau with honeycomb cavities in both patients, indicating abscess formation, and resection was performed. Relatively large cranial FD leads to the development of central necrosis over time. In such cases, surgical intervention should be performed at an early disease stage.

**Key Words:** central necrosis, craniofacial, dysplastic bone, fibrous dysplasia

Fibrous dysplasia (FD) is a developmental, nonfamilial, benign bone lesion characterized by the replacement of normal interior bone tissue by fibro-osseous tissue, which histologically exhibits varying degrees of osseous metaplasia.<sup>1</sup> It is a primary developmental abnormality of the bone-forming mesenchyme in which fibrous tissue gradually expands and replaces bone tissue.

Most craniofacial lesions are asymptomatic. Therefore, surgical resection is typically reserved for symptomatic lesions. Concerning very large lesions, although asymptomatic, surgical resection may be required to improve esthetics. Several surgical experiences have been reported; however, few reports have been published concerning the long-term natural history of FD.<sup>2</sup> In this Brief Clinical Study, we present 2 patients, with implications concerning the natural history of FD and propose earlier surgical intervention in selected cases.

### CASE 1

In August 2023, we followed up a 73-year-old male with a history of right-sided craniofacial FD present since age 8 years. This patient had no wish for surgery despite the lesion having gradually grown to include orbital dystopia (Fig. 1).

On presentation at our hospital, he was febrile with facial inflammation. Computed tomography (CT) findings indicated a massive right-sided craniofacial FD and a niveau with a honeycomb cavity, indicating suspected abscess formation. Owing to the risk of sepsis, emergency drainage surgery, and bone lesion removal were performed. Intraoperatively, the protruding bone was removed with a chisel, and a large amount of pus and necrotic tissue was observed. Complete resection of the bone lesion was not performed, but all honeycomb structures were removed, and the infection was successfully controlled (Fig. 1).

### CASE 2

A 50-year-old male with a history of right-sided cranial FD since childhood presented at our hospital for a thorough examination of the lesion, not having ever visited a hospital previously concerning this FD. He had recently visited a local otolaryngologist, complaining of noise while moving his head. Computed tomography (CT) scans showed niveau in several areas separated by a septal wall (Fig. 1).

The lesion was surgically resected for pathological diagnosis. Most of the accumulated fluid was exudate; however, the