

Temporomandibular Joint Ankylosis In Children

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Abstract: Temporomandibular joint (TMJ) ankylosis is defined as osseous or fibrous fusion of the condyle of the mandible and the mandibular fossa of temporal bone (Nitzan et. al. 1998). It is a debilitating disease that renders the afflicted person unable to use their oral cavity well. Clinical presentation depends on the age at which ankylosis occurs and whether ankylosis is unilateral or bilateral. Surgery and postoperative physiotherapy are regarded as the mainstay treatment for TMJ ankylosis.

The study reviews surgical outcomes of 13 children treated for temporomandibular joint (TMJ) ankylosis in the maxillofacial and oral surgery unit at Red Cross Children's Hospital. Nine patients were treated by gap arthroplasty, while four patients had previous reconstruction with costochondral graft.

One patient treated by gap arthroplasty re-ankylosed. The study showed a positive correlation between intraoperative MIO and the outcome, and between age and the outcome. Mouth opening was improved by 15mm, with postoperative change of -4mm.

Key Words: TMJ ankylosis; maximal interincisal opening; gap arthroplasty; costochondral graft; interpositional arthroplasty and maximal interincisal opening (MIO)

I. Literature Review

Temporomandibular joint (TMJ) ankylosis is defined as osseous or fibrous fusion of the condyle of the mandible and the mandibular fossa of temporal bone¹. TMJ ankylosis is a rare condition that renders the afflicted individual unable to masticate, articulate well and maintain good oral hygiene^{2,3,4,5}. Some patients suffer from obstructive sleep apnoea due to small retruded mandible⁶. This condition deprives the afflicted person of good nutrition and opportunity to have proper dental treatment^{3,2}. Inability to remove exfoliated teeth in children with TMJ ankylosis has been reported as well in patients with TMJ ankylosis⁷.

The incidence of TMJ ankylosis differs significantly in different parts of the world. Studies from Egypt^{8,4,9} and India¹⁰ present with large samples, while studies from other parts of the world report fewer number of patients seen over a long period^{11,12,13,14}. Kazanjian (1938) classified TMJ ankylosis according to the site involved into true (intracapsular) and false (extracapsular) ankylosis. Intracapsular ankylosis refers to fibrous or bony ankylosis that occurs between the condylar head of the mandible and the mandibular fossa of the temporal bone. While extracapsular ankylosis refers to restriction of mandibular movement that occurs as result of pathology or physical obstruction that is outside the TMJ¹⁰. Intracapsular (True) ankylosis was further classified according to extent by Topazian and Sawhney^{15, 16}.

Aetiology of TMJ ankylosis may either be congenital or acquired. Acquired TMJ ankylosis may be caused by trauma, infection^{11,16,4,2}, systemic inflammatory disorders¹⁷, irradiation, previous surgery^{2,18} and neoplasm¹⁹. The posttraumatic TMJ ankylosis follows misdiagnosis, delayed treatment, inadequate surgery, prolonged immobilisation or insufficient physiotherapy²⁰.

Most studies suggest that trauma is the most common cause of TMJ ankylosis^{8,20,12,6}. In the pre-antibiotic era infection was the most common cause of ankylosis²¹. However Kaban et al 2009 maintain that infection is still the most common cause of ankylosis in the third world countries¹². Some studies suggest that disc displacement is a prerequisite for posttraumatic ankylosis to occur. These authors maintain that this displacement allows a direct contact between the distal fragment of the mandible and the mandibular fossa or a clot occupies the region between the two surfaces^{15,1}.

In children clinical presentation depends on the age at which ankylosis occurs, duration and on whether ankylosis is unilateral or bilateral^{22,12}. When ankylosis occurs after growth has ceased, it presents mainly with limitation of mouth opening^{21,20}. Currently, the surgical techniques used to treat TMJ ankylosis are gap arthroplasty, interpositional arthroplasty, joint reconstruction²⁴ and distraction osteogenesis^{5,18}. Some authors prefer to correct secondary deformities like facial asymmetry, occlusal canting and micrognathic mandible concomitantly with the release of the ankylosis^{22,8,13}.

The goals for the release of TMJ ankylosis are to create a pseudoarthrosis that will improve function or movement of the mandible^{21,20,6}, prevent relapse²¹, relieve airway obstruction if present^{8,2}, achieve normal growth and correction of deformity in children^{6,12}, restore appearance and occlusion in adults²¹ and facilitate maintenance of good oral hygiene.

Reankylosis is the most common complication after surgery²⁴. The reported causes of reankylosis are inadequate removal of ankylosic mass^{25,26} and lack of compliance to post-operative exercises^{20,27}. Other complications that have been reported include profuse bleeding, transient facial nerve paralysis^{4,28} and sepsis⁴. While the following complication associated with use of costochondral graft are fracture of the costochondral graft (CCG)²⁹, overgrowth of the graft³⁰ and suboptimal growth³¹. Anterior open bite occurred in patients treated with gap arthroplasty²⁸.

Lello (1990) in his series of 13 cases that were treated by condylectomy, reconstruction with costochondral graft and use of interpositional material, only one case reankylosed³².

Guyuron and Lasa (1992) reported an unpredictable behaviour of costochondral graft, in 8 patients between 4 and 12 years, who had reconstruction with autogenous costochondral graft. In 4 patients there was excessive growth of the graft, 1 patient had sub-optimal growth and 3 patients had no growth³³.

Manganello-Souza and Miriani (2003) reviewed 14 patients who were managed by interpositional arthroplasty. Recurrence occurred in one patient (7.1%)²³.

El-Mofty (1974) presented his results of 20 cases with unilateral TMJ ankylosis. Seventeen of these cases were 12 years or younger. All patients were treated by gap Arthroplasty. Recurrence occurred in 2 patients (10%)⁴. Rajgopal et.al. (1983) achieved 100% success with gap arthroplasty²⁸. Roychoudury et.al. (1999), also reported good results of 50 cases treated with gap arthroplasty. Mouth opening was improved in both children and adults, and reankylosis occurred in one patient (2%)³³.

These studies show that reankylosis is common complication irrespective of surgical technique used to treat the TMJ ankylosis.

The objective was to evaluate outcomes in children treated for TMJ ankylosis at Red Cross Children's hospitals; and to establish factors that influence the outcomes in Children.

II. Materials And Methods

Study Design

This is a retrospective study of the records of 13 patients treated for TMJ ankylosis in the Maxillofacial and Oral Surgery unit at Red Cross Children's Hospitals (RXH). All patients operated for TMJ ankylosis from January 1988 to March 2003 were included in this study.

Surgical Data

All patients were treated by the protocol described below:- Patients had Plain x-rays and computated tomography as standard imaging for confirming diagnosis of TMJ ankylosis. Preauricular incision was made. Excision of ankylosic mass. If adequate mouth opening was not achieved, ipsilateral coronoideectomy was done. Contralateral coronoideectomy via intra-oral approach (where necessary). When reconstruction was planned, costochondral graft was used. Modified submandibular incision was made. Stripping of the masseter from its insertion was done. Harvest of costochondral graft from rib 5, 6 or 7. Costochondral graft was stabilised screws on the lateral aspect of the ramus of the mandible. Maxillary and mandibular alginate impressions were taken after the release of TMJs. After the operation mouth prop was used to keep the patients in maximum interincisal opening for 24hrs. Exerciser was delivered 24hrs after surgery.

Data Collection

Clinical records were retrieved from RXH. Patients who were 12 years and younger during surgery were be grouped as pediatric cases. Only patients with intracapsular ankylosis were included in the study. A ruler was used to measure maximal interincisal opening (MIO). The data that were analysed were categorised into pre-operative, intra-operative and post-operative phases. Preoperative data were analysed for age of the patient at the first visit, sex, aetiology, whether the ankylosis was unilateral or bilateral, type of ankylosis (fibrous or bony) and pre-operative maximal interincisal opening (pre-op MIO). Operative data included type of surgery, achieved intraoperative maximal interincisal opening (Op MIO) and intubation technique used. Postoperative data were reviewed for the outcome MIO (the last recorded maximal interincisal opening), complications and follow up period. In patients that were operated more than once, the data from the last operation was used.

In this study the outcomes were evaluated with reference to two parameters, namely pre-op MIO and Op MIO. Each of these variables was subtracted from the outcome MIO. A variable that uses pre-op MIO as a reference point is called an overall change in MIO (OC MIO) and one which uses Op MIO is called post-operative change in MIO (Pop MIO). OC MIO of 10mm or more was regarded as good outcome. OC MIO less than 10mm was regarded as reankylosis.

Ethical issues

The study was approved by Research Committee of the University of Western Cape. Permission to use hospital records was given by the hospital manager. Strict confidentiality was adhered to. Numbers were used to identify patients.

Analysis of results

Tables and graphs were used to demonstrate the outcomes. Statistica version 12 was used for statistical analysis of results.

III. Results

The age ranged between 4 years and 12 years. Seven of the patients were females and 6 were males. The aetiology of ankylosis was congenital, trauma and infection in 7, 4 and 2 patients respectively. However in two patients that were thought to have a congenital cause of ankylosis, a condyle of the mandible was found lying on the medial aspect of the ankylosis mass. This was suggestive of the trauma as the cause of ankylosis.

patient	Age	Sex	Aetiology	Side	Type	Airway	Preop MIO (mm)
2.	6	Male	Tr (mva)	Unilateral	Fibrous	Competent	8
4	5	Male	Congenital	Bilateral	Fibrous	Competent	8
6.	12	Female	Congenital	Unilateral	Bony	Competent	20
9.	4	Female	Congenital	Unilateral	Bony	Tracheotomy Dependant	0
10	5	Female	Congenital	Bilateral	Bony	Tracheostomy Dependant	14
11.	10	Male	Congenital	Bilateral	Fibrous	Tracheostomy Dependant	10
18	9	Male	Congenital	Bilateral	Bony	Competent	1
22	12	Female	Tr. (mva)	Unilateral	Bony	Competent	5
27.	6	Male	Infection	Bilateral	Bony	Competent	0
28.	5	Female	Infection	Bilateral	Bony	Competent	0
29.	12	Male	Congenital	Bilateral	Bony	Competent	0
36	8	Female	Trauma	Unilateral	Fibrous	Competent	15
45	4	Female	Trauma	Bilateral	Bony	Tracheostomy Dependant	8

Table 1 shows preoperative data

TMJ ankylosis was unilateral in 5 patients and bilateral in 8 patients with total of 21 ankylosed joints. Four patients were tracheostomy dependant and 3 of these patients had bilateral TMJ ankylosis. Nine patients with intracapsular ankylosis were treated by gap arthroplasty, while four had grafting of the joint with costochondral graft. Both blind nasal intubation and fibre optic intubation were used in two and four patients respectively. While one patient was intubated by direct laryngoscopy.

Most patients had a negative postoperative change. This suggests that Op MIO was reduced, while few patients had a positive Pop MIO. Two patients re-ankylosed, both patients had unilateral bony ankylosis and were managed by gap arthroplasty. These two patients had a poor Op MIO of 15 mm and 20 mm.

Case	Outcome MIO (mm)	OC MIO (mm)	Pop MIO (mm)	MIO (mm)	Complication	Follow Up (months)
2	33	25	8	None	37	
4	33	25	5	None	61	
6	27	7	-13	Re-ankylosis	48	
9	0	0	-20	Re-ankylosis	36	
10	26	12	-4	None	66	
11	25	15	-2	None	15	
18	26	25	-2	None	60	
22	31	26	6	None	24	
27	10	10		None	120	
28	10	10	-5	None	132	
29	22	22	-12	None	48	
36	34	19	-1	None	106	
45	25	17	-5	None	140	

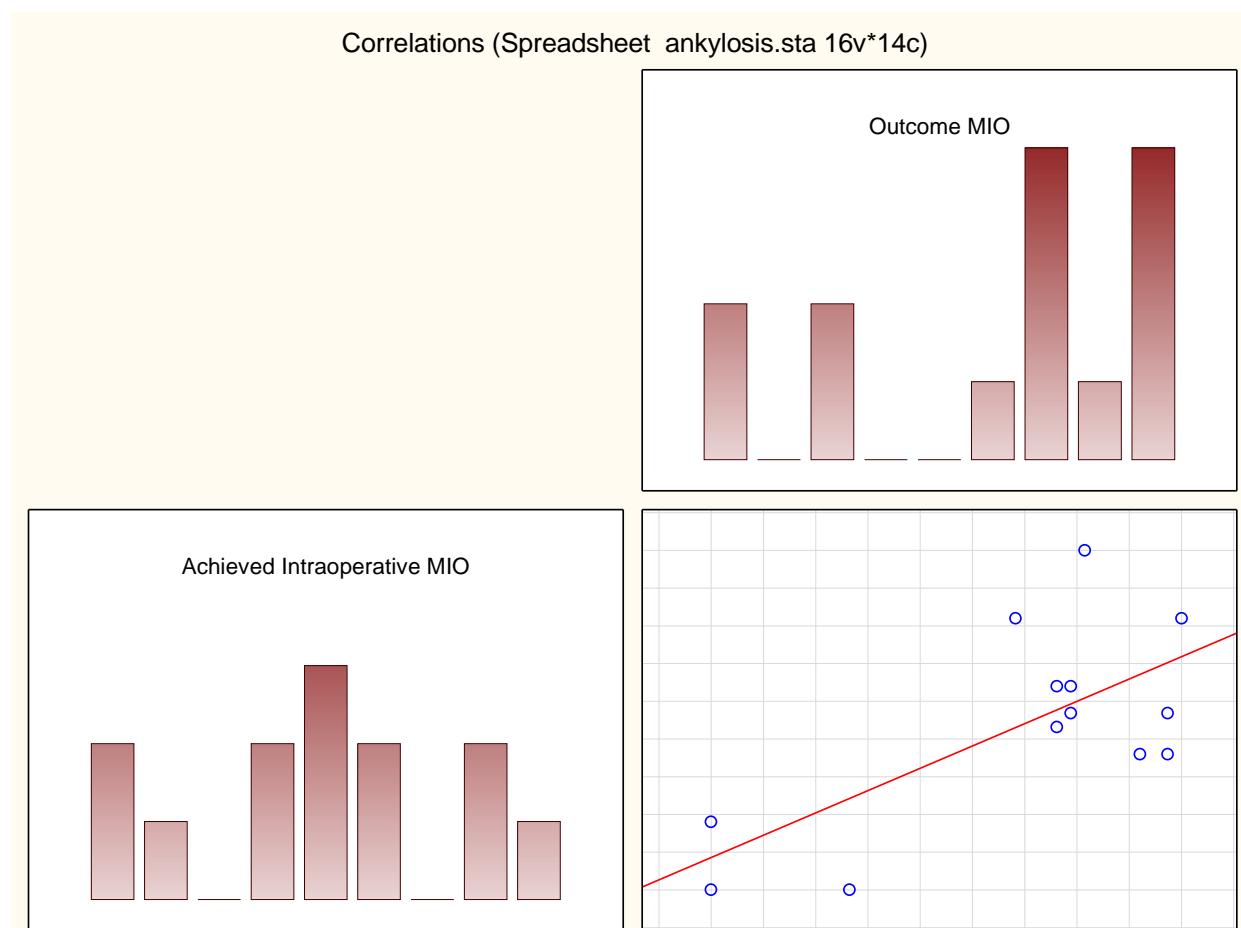
Table 2 shows outcomes in the series

IV. Discussion

The study showed that aetiology was fairly distributed between trauma and congenital cases. Although the history suggested that congenital cases were more common, two cases with unilateral TMJ ankylosis whose aetiology was congenital according to history given, proved to be traumatic as the condyle was retrieved on the medial aspect of the ankylosic mass during surgery. This finding suggests that a **unilateral congenital** TMJ ankylosis should be confirmed during surgery by absence of a condyle. This finding supports the view of some authors who believe that the term "congenital ankylosis" tends to be applied to patients who had limitation of mouth opening at infancy which might have been caused by trauma during delivery^{15,28}. However, El-Sheik (1999) applies the term congenital ankylosis to a condition where there is complete bony fusion between mandibular condyle and temporal bone, with absence of any trace of intra-articular disc and lateral pterygoid muscle⁸. This series showed that all congenital cases had bilateral TMJ ankylosis. This concurs with findings of Posnick and Goldstein (1993)³⁴.

Four patients in this series were tracheostomy dependent and three of these patients had bilateral TMJ ankylosis. TMJ ankylosis in these tracheostomy-dependent patients was either congenital or occurred in the first year of life. The airway compromise in these patients is due to relative normal growth of the tongue, within a confined space which is bounded by slow or non-growing mandible, resulting in the posterior displacement of the tongue^{35,8}. This suggests that bilateral TMJ ankylosis that occurs very early in life is more likely to result in a compromised airway.

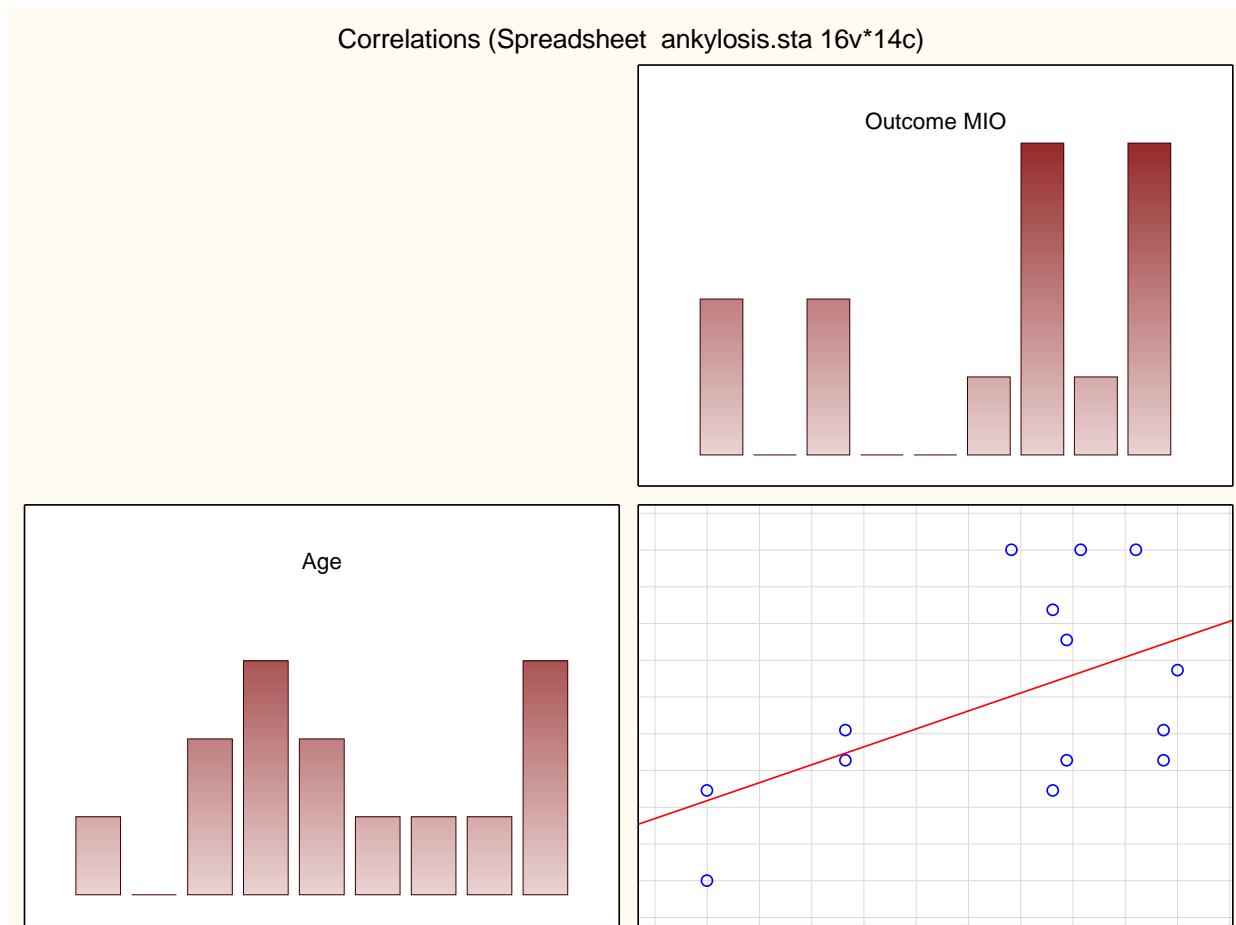
This series shows positive correlation between intraoperative MIO and the outcome MIO according to Spearman R (*ref graph 1*). This suggests that good Op MIO is one of the important factors that influence the outcomes in TMJ ankylosis. This concurs with Kaban et al 2009 who maintain that inadequate release of ankylosic mass is the main cause of reanklylosis¹².



Graph 1: Scatter plot graph shows correlation between the intraoperative MIO and the Outcome

When the postoperative data was analysed, a mean postoperative progress shows a varying reduction of what was achieved intraoperatively with a mean of -4,6 mm. However the study shows a mean overall change of 15,2 mm.

The study also showed a **positive correlation** between the age and the outcome. This correlation was statistically significant with $p<0,05$ (ref graph 2). The explanation for this positive correlation is that older patients understand the objective of the treatment, are more likely to cooperate with the exercises, resulting in good outcomes. In this series four patients had been previously reconstructed with costochondral graft and re-ankylosed. These patients were in their first decade during reconstruction. When reconstruction with costochondral graft was done for the second time, good results were obtained. It is assumed that these good results are achieved because patients in their teens understand the impact of the deformity and compliance to physiotherapy is better. This finding also supports authors who maintain that reconstruction with costochondral graft should be a secondary procedure after gap arthroplasty^{37,39}.



Graph 2: Scatter plot graph showing correlation between age of the patient and outcome

Literature suggests that costochondral graft (CCG) is a material of choice for reconstruction of TMJ in children³¹. The advantages of CCG is that it offers three important functions ; cartilagenous portion act as an interpositional material²⁴, the bony portion restores the vertical height of the ascending ramus²⁴ and the junction between cartilaginous portion and the bony portion act as a growth centre^{36,35}. The main objective of using costochondral graft in children is to transplant a growth centre^{36,34}. The disadvantages of CCG are second operation site and donor site morbidity²⁶ and unpredictable growth of the costochondral graft has been reported. Costochondral graft growth may be normal, suboptimal or may overgrow³¹.

These outcomes with costochondral graft concur with some studies that have reported poor results with costochondral graft³⁸. Guyuron and Carlos (1992) reported unpredictability of costochondral graft as the main disadvantage because in their series of eight cases, overgrowth occurred in 4 cases, no growth in three patients and sub-optimal growth in one patient³¹. However, Lello (1990) reported good surgical outcomes with costochondral graft and interpositional material³².

Review of the literature shows that outcomes are mainly assessed by using the final IID, whereby outcomes are grouped into ranges of IID of 10-20mm; 20-30mm and 30 and above¹⁵. Chessgros et al (1997) regard an IID of least 30mm as a good result³⁵, while Kaban et al (1990) believe an IID of 35mm is a good outcome²⁶. However, Topazian (1966) regards an IID of 20mm as adequate for function²⁵.

Variable	No of cases	Mean	minimum	Maximum	STD Deviation
Pre-op MIO	13	6.4	0.0	20	6.6
Op MIO	12	27	15	40	7.4
Outcome MIO	13	21.6	0	34	11.8
Post op MIO	13	-4.6	-20	8	8.5
OC MIO	13	15.2	0	26	9.0
Follow up	13	67	15	140	40

Table 4 shows descriptive statistics of the outcomes of the study

This study introduces a new parameter in the study of outcomes in patients with TMJ ankylosis. This parameter, post-operative change in MIO (Pop MIO), actually shows the progress after surgery. It measures the actual impact of physiotherapy on the outcomes. Pop MIO can also be used in motivating and encouraging the patient to continue with the exercises and can also be used as a research tool. A positive Pop MIO should be a goal of every patient. The main disadvantage of this variable is that it cannot be worked out if intra-operative MIO was not recorded.

In the literature there is an agreement on the importance of aggressive physiotherapy after the release of TMJ ankylosis, but there is still controversy on when to start with the mobilisation of the jaws. Some authors believe in early mobilization ^{17,33,28}, while others immobilise the patient for a period of 1-10 weeks^{26,34,8}. In this series an appliance was used for exercises.

The mean follow up time was 67 months with range of 15 to 140 months.(table 4)

V. Conclusion

This study suggests that there is positive correlation between preoperative MIO and intraoperative MIO and the outcome MIO.

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