Temporomandibular Joint Ankylosis in Children: Five Year Experience from a Tertiary Care Institute

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Abstract

Aim: Temporomandibular joint ankylosis in children develops following unnoticed trauma to the chin. In chronic cases, a group of complex deformities develop which pose significant challenges in its management. Surgery followed by physiotherapy are pivotal steps for its management and for prevention of recurrence. In this study, we aimed to highlight our experiences with eleven cases of temporomandibular joint ankylosis which were managed successfully in our institution during last 5 years.

Materials and Methods: We analyzed the surgical outcomes of interpositional arthroplasty of 11 children treated for temporomandibular joint ankylosis over a period of five years. The specifics of all patients in the form of patient's demography, preoperative assessment, perioperative findings and follow-up assessment were recorded, tabulated and analyzed carefully.

Results: Ten patients required tracheostomy intubation for general anesthesia. No difficulty was faced during tracheostomy and its closure. Standard incision followed by intersessional arthroplasty using temporalis muscle was done in all cases. Operative success were assessed by comparing the preoperative and postoperative maximal incisal opening after 6 weeks and by 2 years.

Conclusion: Osteoarthotomy with interposition arthroplasty is most viable option for the management of all grades of temporomandibular joint ankylosis in children. Severe ankylosis with maximal incisal opening less than 8 mm can safely be intubated with tracheostomy. Postoperative aggressive physiotherapy under supervision is imperative to procure the maximal functional outcome.

Keywords: Ankylosis; Temporomandibular Joint; Children; Tracheostomy; Arthroplasty; Physiotherapy

Introduction

Temporomandibular joint ankylosis (TMJA) is a pathological term that means fusion of the articular surfaces of the condyle of the mandible and the mandibular fossa of temporal bone with bony or fibrous tissue. Trauma seems to be the most common harbinger of TMJA. The condition usually presents with difficulties in feeding, mastication, speech impairment, facial deformity with psycho-social noncooperation. Severity of symptoms depend on the duration of the disease, the age at which the patient presents, unilateral or bilateral and the range of motion of the mandible [1]. Prime aim of management is to restore joint mobility and prevention of reankylosis. Surgery, which

has evolved from a gap arthroplasty to osteoarthotomy with interposition arthroplasty with temporalis myofascial flap or acrylic spacer followed by aggressive physiotherapy are imperative to manage the disease as well as for prevention of re-ankylosis [2].

**Materials and Methods**

Eleven cases of TMJA were retrospectively reviewed. The children below 12 years who underwent interposition arthroplasty using temporalis muscle (OIAT) for TMJA in the Department of Pediatric Surgery in a tertiary care center from April 2014 to March 2018 were included in this study. All cases had a history of trauma to the chin. CT of TMJ was performed in all patient in addition to blood hemogram and blood biochemical examinations as preoperative investigation. The indications of surgical intervention in our study were maximal incisal opening (MIO) less than 1.4 cm or less, feeding difficulty and poor oral hygiene.

**The procedure of OIAT through preauricular approach**

Patient is placed in supine position and a transverse incision is made 2.5 above the suprasternal notch for tracheostomy under local anesthesia with sedation. After tracheostomy, an appropriate sized cuffed tube is placed through tracheostomy for administration of general anesthesia. Then patient’s head is tilted to opposite side to make the operative side accessible. For OIAT, Al khayat branley’s incision is used. A question mark shaped preauricular incision extending up to the temporal region is applied. Then the incision deepened to the depth of the temporalis fascia in the scalp and the temporalis muscle is exposed up to the pinna. The incision is then extended antero-inferiorly up to the ear lobule. Then blunt dissection is carried out in an avascular plane till the condylar head is palpable. It is done carefully to not injure the branches of facial nerve and perforation of the articular cartilage. Then, root of the zygoma is palpated in the temporal region and about 3 - 4 cm incision is made to the depth of the bone making an angle of 45 degree to the zygomatic arch. A periosteal elevator is introduced through the incision and dissection is continued in an inferior direction towards condylar head in a back and forth manner till the capsule of the TMJ is seen. The capsule of the TMJ is incised with the scalpel blade and dissection continues inferiorly for about 1 - 2 cm below the condylar head. Now with the periosteal elevator over the neck of the condyle, incision is completed over the preauricular region to expose the condyle and the sub-condyle. Temporalis muscle is stripped from the coronoid process with the periosteal elevator and the whole ankylotic mass is exposed. Osteotomy is done to create a gap of 1 - 1.5 cm keeping a periosteal elevator on the medial aspect of the neck of the condyle to prevent injury to the internal maxillary artery and pterygoid venous plexus. A slip of temporalis muscle with fascia is elevated, rotated and placed in the gap and secured with sutures to act as an interposition materials. Inter-incisional opening is assessed and if needed opposite site is operated in similar fashion. The wound is closed in layers after placement of corrugated rubber drain. Postoperative analgesia was provided with paracetamol.

Post-operatively all patients is sent to PICU for ventilatory support. Tracheostomy tube extubation is initiated from 3rd post-operative day (POD) onwards. Oral feeding is started on 4th POD. Post-operative physiotherapy is started once the patient starts to take full meal and it is continued for at least 6 months. All the patients are followed up for 24 months to note the psychosocial improvement and any clinico-radiological evidence of recurrence. A scrutiny for transient facial nerve paralysis and serial measurement of maximal incisal opening are noted carefully in follow-up clinic. Detailed data of patient’s demographical profile, age at operation, preoperative clinical data, reports of imaging studies, operative finding, details of surgical technique, operating time, need of blood transfusion, time required for resumption of oral intake, hospital stay, and postoperative complications, pre and post-operative maximal incisal opening, and post-operative follow-up records were collected and tabulated and analyzed (Table 1).
### Table 1: Clinical spectrum and Surgical outcome of temporomandibular joint ankylosis (n = 11).

<table>
<thead>
<tr>
<th>SL No</th>
<th>Gender</th>
<th>Age at Diagnosis</th>
<th>Age at Operation</th>
<th>Involvement</th>
<th>CT of TMJ</th>
<th>MIO Pre-op</th>
<th>Elongation of coronoid</th>
<th>OT</th>
<th>Tracheostomy</th>
<th>EXT</th>
<th>BT</th>
<th>Complications</th>
<th>Hosp. Stay</th>
<th>MIO at discharge</th>
<th>MIO at 6 months</th>
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<tr>
<td>1</td>
<td>M</td>
<td>4 Yr</td>
<td>5 Yr</td>
<td>L</td>
<td>T3</td>
<td>12 mm</td>
<td>No</td>
<td>2 hr</td>
<td>Yes</td>
<td>4POD</td>
<td>0</td>
<td>No</td>
<td>11D</td>
<td>34 mm</td>
<td>36 mm</td>
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<tr>
<td>2</td>
<td>F</td>
<td>4 Yr</td>
<td>7 Yr</td>
<td>L</td>
<td>T3</td>
<td>10 mm</td>
<td>Yes</td>
<td>3 hr</td>
<td>Yes</td>
<td>4POD</td>
<td>1 Unit</td>
<td>No</td>
<td>14D</td>
<td>35 mm</td>
<td>35 mm</td>
</tr>
<tr>
<td>3</td>
<td>M</td>
<td>5 Yr</td>
<td>6 Yr</td>
<td>R</td>
<td>T3</td>
<td>14 mm</td>
<td>No</td>
<td>2 hr</td>
<td>No</td>
<td>4POD</td>
<td>0</td>
<td>No</td>
<td>14D</td>
<td>35 mm</td>
<td>36 mm</td>
</tr>
<tr>
<td>4</td>
<td>F</td>
<td>4 Yr</td>
<td>6 Yr</td>
<td>L</td>
<td>T3</td>
<td>8 mm</td>
<td>Yes</td>
<td>4 hr</td>
<td>Yes</td>
<td>5POD</td>
<td>2</td>
<td>No</td>
<td>14D</td>
<td>33 mm</td>
<td>34 mm</td>
</tr>
<tr>
<td>5</td>
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<td>5 Yr</td>
<td>8 Yr</td>
<td>R</td>
<td>T3</td>
<td>8 mm</td>
<td>No</td>
<td>2.3 hr</td>
<td>Yes</td>
<td>4POD</td>
<td>0</td>
<td>No</td>
<td>12D</td>
<td>32 mm</td>
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</tr>
<tr>
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<td>B/L</td>
<td>T4</td>
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<td>Yes</td>
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<td>No</td>
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<td>Yes</td>
<td>4POD</td>
<td>0</td>
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<td>12D</td>
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<tr>
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<td>R</td>
<td>T4</td>
<td>3 mm</td>
<td>No</td>
<td>2 hr</td>
<td>Yes</td>
<td>4POD</td>
<td>1 Unit</td>
<td>No</td>
<td>19D</td>
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<tr>
<td>9</td>
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<td>T4</td>
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<td>Yes</td>
<td>2.3 hr</td>
<td>Yes</td>
<td>7POD</td>
<td>0</td>
<td>No</td>
<td>11D</td>
<td>33 mm</td>
<td>34 mm</td>
</tr>
<tr>
<td>10</td>
<td>M</td>
<td>6 Yr</td>
<td>9 Yr</td>
<td>L</td>
<td>T3</td>
<td>10 mm</td>
<td>Yes</td>
<td>1.3 hr</td>
<td>Yes</td>
<td>5POD</td>
<td>1 Unit</td>
<td>No</td>
<td>14D</td>
<td>36 mm</td>
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<tr>
<td>11</td>
<td>M</td>
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<td>L</td>
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<td>8 mm</td>
<td>Yes</td>
<td>2.3 hr</td>
<td>Yes</td>
<td>4POD</td>
<td>1 Unit</td>
<td>No</td>
<td>14D</td>
<td>34 mm</td>
<td>36 mm</td>
</tr>
</tbody>
</table>

**Abbreviations:** MIO: Maximal Incisal Opening; T3/T4: Sawhney’s Classification Type 3/Type 4; OT: Operative Time; BT: Blood Transfusion; U/L: Unilateral; B/L: Bilateral; EXT: Tracheostomy Tube Extubation; *Complication: Open Site with Drooling of Saliva.

**Figure 1:** Pictures of computed tomography and maximal incisal opening in a case of left sided ankylosis.
Results

A total of 11 patients with TMJA were enrolled into this study. Of them, 5 (45.45%) were male and 6 (54.54%) were female patients. They were divided into two groups on the basis of the side of the aortic arch. Ten patients (90.90%) had unilateral and one patient had bilateral ankylosis. There was wide gap with drooling of saliva in just one bilateral case. Out of 10 unilateral ankylosis, left side (70%) was more predominant than the right side (30%). The mean age at the time of operation was 6.81 years (range 4 to 10 years). Preoperative CT scan revealed various types of ankylosis, according to Sawhney’s classification type III (n = 8) and type IV (n = 3) and that diagnosis was confirmed intra-operatively. OAIT was done in all patients using temporal fascial flap. There was no difficulty in gaining the access to the pathology in any of the patients even in BL case. Resection of coronoid process was undertaken in 6 cases (54.54%). Pre-operative MIO range from 3 mm to 14 mm with a mean of 8.36 mm which was increased to a mean of 34 mm (range 31 to 36 mm) at the time of discharge and 35.18 mm (range 33 - 38 mm) after 6 month. General anesthesia was administered with tracheostomy intubation in ten cases because of too small mouth opening. All patients recovered from anesthesia uneventfully and returned to full oral feeds by 4th postoperative day. The mean time of tracheostomy tube extubation was 4.72 days (range 4 days to 7 days). There were no major complications related to surgery or tracheostomy tube extubation. Temporary facial nerve paralysis was encountered in one patient (9.09%) which resolved subsequently within a week. Five patients (45.45%) required blood transfusion in postoperative period. The mean duration of hospital stay was 12.27 days with a range of 11 days to 19 days. The average length of incision was 3 - 4 cm and mean operating time was 156 minutes with a range of 80 minutes to 260 minutes. No recurrence was observed in our series. No operative or disease related death occurred in our series.

Figure 2: Intra-operative pictures of interposition arthroplasty using temporalis muscle.
Discussion

The word ankylosis means stiff joint. TMJA is characterized by restricted mouth opening because of fusion of joint surfaces by bone or fibrous tissue. Unlike western countries, India has still high incidence of TMJA. Most common cause is unnoticed trauma to the chin. However, TMJA may occur following an infection of the joint [3]. It is a progressive and enervating disease that affects the normal growth, development of jaws and occlusion. Pathophysiological basis is excessive mineralization and bone formation with comminuted intracapsular fractures due to prolonged immobilization leading to fibrous or bony union [4]. It may be intra or extra-articular; complete or incomplete (depending on the degree of fusion) and bony, fibrous or fibro-osseous [5]. In unilateral disease, chin deviates to the diseased site (lateral displacement of the chin) with flattening of unaffected site. Because of progressive trismus, there will be poor dental health and oral hygiene with multiple decayed teeth, difficult chewing, poor nutrition, growth stunting, and impairment of speech. In long standing case, severe facial deformity like; “Bird’s facies” retrognathia and obstructive sleep apnoea develops especially if the disease process starts early in growing age [6]. Inability to open the mouth becomes a mental stigma especially in older children. Diagnosis of TMJA is purely clinical. However, certain clinical and radiographic evidences must be considered for more precise diagnosis and surgical planning. To determine the extent of the ankylosis panoramic radiographs and axial as well as coronal computed tomography (CT) are sufficient. Sawhney’s classification was developed on the basis of CT scan findings. Type I: minimal bony fusion, but extensive fibrous adhesions around the joint. Type II: bony fusion in the outer edge of the joint surface, but no fusion within the more medial area of the joint [7]. Type III: a bridge of bone between the mandible and the temporal bone and type IV: the joint was replaced by a mass of bone. Fibrous ankylosis can be distinguishable from bony ankylosis by two ways. Clinically, patient will be able to open mouth forcefully and protrude the mandible slightly. Radiologically, patient has a narrow joint space and appearance of jigsaw puzzle (two erotic and irregular surfaces appear to fit one another like a jigsaw puzzle).

Surgery followed by physiotherapy is the mainstay treatment to restore joint function and prevent re-ankylosis for TMJ ankylosis. Three main surgical techniques are described in literature. (1) Gap arthroplasty (GA), Resection of the osseous mass between the articular cavity and the mandibular ramus. (2) Interpositional arthroplasty (IA) evolving as OIAT now, placement of interpositional material in the gap created by resecting the osseous mass. (3) Reconstruction arthroplasty (RA), after complete resection of the osseous mass and joint reconstruction by autogenous bone grafts or joint replacement by total joint prosthesis [8,9]. Two surgical techniques (GA and OIAT) are currently practiced by orthodontic surgeons. GA is technically less demanding and oldest surgical technique for TMJA which required shorter operative time than other two procedure [10,11]. OIAT has been evolved to reduce the occurrence of re-ankylosis by insertion of interpositional materials after resection of the ankylotic mass [12,13]. In our series, we performed OIAT in all patients and got a fruitful result. The procedure of OIAT is nicely described in Success in Kaban’s protocol. It is consist of following steps 1. Aggressive resection of ankylotic mass, 2. Ipsilateral/contralateral coronoidectomy, 3. Placement of interpositional materials and 4. Early mobilization with aggressive physiotherapy [14,15]. All the graft materials have their own limitations such as: a muscle shrinks, a fascia lacks bulk, a cartilage gets calcification, an uncontrolled growth of costochondral graft leads to asymmetric facial profile and overgrowth in TMJ region and an alloplastic implant or nonbiological material (acrylic, silastic) tends to produce foreign body reactions. Topazian advised to create a partition between the two resected surfaces (“functional pseudoarthrosis) using an autologous materials. Though, a few comparative studies with GA and OIAT do not specifically said that OIAT is superior over GA [16,17].

Administration of anesthesia to these patients also create a dilemma. As, endotracheal intubation becomes very tough/impossible because of narrowed mouth opening, lack of direct visualization of the vocal cords and associated retrognathia. Various integrated intubation techniques have been evolved to overcome these challenges. Like; blind nasal intubation, fiber optic laryngoscope-assisted intubation, bi nasopharyngeal airway, fluoroscope-aided retrograde placement of guide wire for tracheal intubation, retrograde endotracheal intubation using a pharyngeal loop and semi blind technique of nasal intubation [18]. However, blind nasal intubation is discouraged because of high failure rates and risk of trauma to the air passage. Intubation with Fibre optic laryngoscope requires a set of costly endo-
scopes of various sizes [19]. Gaiiwala., et al. reported a case of TMJA where surgery was performed under local anesthesia successfully but it may not be adequate in all cases of TMJA [20]. In our series, 10 patients underwent tracheotomy intubation for OIAT. We believe, Tracheostomy is a good and safe option for severe form of TMJA. Moreover, maintenance of airway in postoperative period becomes easier. We did not encounter any problem with tracheostomy extubation and early initiation of oral feeding.

Truly, surgical correction is technically difficult in type IV ankylosis in children. And, prevention of recurrence after treatment is more challenging. An inadequate resection of the ankylotic mass and failure to achieve adequate passive maximal opening during surgery are the most important determining factors for treatment failure [21]. Kaban., et al. reported that an inadequate release of ankylotic mass is the main cause of re-ankylosis. Sometimes, to achieve adequate passive maximal opening during operation contralateral coronoidectomy is needed. To reduce the recurrence rate, OIAT or JA procedure and aggressive physiotherapy in the immediate post-operative period are imperative. Success of surgical management is usually assessed in the postoperative period in the form of assessment of mouth opening, status of occlusion, facial symmetry and radiographic evaluation of graft position, joint form, function and bilateral symmetry [22,23]. In our series, we followed-up all the cases and got satisfactory occlusion and acceptable facial symmetry as well as mandibular function. Interestingly, we found a positive correlation between the age and the outcome. The result is good in older children because they are more cooperative in postoperative physiotherapy as they better understand the impact of the deformity and compliance to physiotherapy.

Conclusion

As far as our small series is concerned OIAT procedure is better than other conventional techniques bearing a success rate of 100% in the management of TMJA. It is also successful in majority of the type IV cases of TMJA. One of the limitations of our study may be the lack of comparison group to GA or RA. Another limitation is restricted number of cases (n = 11). Considering our result and recent literatures, we warrant that specific technique based on institutional practice, proper counselling of both patient and their parents and a psychological rehabilitation have a positive impact in the management of TMJA.

Bibliography


