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Management of paediatric ankylosis



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ABSTRACT

Temporomandibular joint ankylosis (TMJa) is one of the most crippling craniomaxillofacial pathological conditions characterized by replacement of normal architecture of temporomandibular joint (TMJ) with fibrous or bony tissue. The incidence of TMJa is most common in the paediatric population [first and second decades of life] and is commonly associated with maxillofacial trauma. Comprehensive management entails a thorough evaluation of the associated anatomy of the ankylotic mass and other pertinent details like the presence or absence of obstructive sleep apnoea. Categorizing patients based on these variables helps in selecting an appropriate surgical intervention. Various resective and reconstructive surgical techniques are discussed; along with their merits and demerits. Long-term physiotherapy, long-term clinical follow-up and appropriate family counselling are the essential pillars for success. In this review, the authors present an algorithmic approach to evaluation and management of paediatric TMJa. Appropriate recommendations are made based on evidence to select optimum surgical intervention.

1. Introduction

Temporomandibular joint ankylosis (TMJa) is one of the most crippling craniomaxillofacial pathological conditions characterised by replacement of normal architecture of temporomandibular joint (TMJ) with fibrous or bony tissue. The incidence of TMJa is most common in first and second decades of life (35–92%), with males being more commonly affected than females. It is most commonly associated with trauma (13–100%), local or systemic infection (10–40%), systemic diseases (10%) such as ankylosing spondylitis, rheumatoid arthritis, psoriasis and previous TMJ ankylosis surgery.^{1–3}

Features of TMJa can be divided into two main sub-groups: limited range of TMJ motion and limited or deviated mandibular growth. A limited range of TMJ motion leads to difficulties in mouth opening, masticatory efficiency, speech and oral hygiene. Limited or deviated mandibular growth presents as mandibular micrognathia, facial skeletal asymmetry, malocclusion and sleep apnoea [Figs. 1 and 2]. On account of its debilitating presentation, TMJa aggravates psychological stress and severely decreases the patient's quality of life.^{1–3}

This review aims to present the current recommendations for the management of paediatric TMJa. Primary and secondary reconstruction of the TMJ will be covered in detail in separate sections of this issue.

2. Aetiology and genetic expression in TMJ ankylosis

TMJ ankylosis usually results from injury (13–100%), local or systemic infection (10–40%), or systemic disease (10%), such as ankylosing spondylitis, rheumatoid arthritis, and psoriasis. Other reasons are iatrogenic causes such as cytotoxic medication, repeated TMJ surgery and irradiation.¹⁷

In recent times, many genetic expression studies have been conducted to determine if there is a genetic predisposition for TMJa. A pilot study conducted by André da Silveira Braune in 2020, showed low RANKL expression in the test group; when compared with the control group. This may be evidence of a possible change in the bone homeostasis of children with TMJa that favours bone synthesis.⁵⁶

Paola Fernanda Corso also conducted a study in 2022 to find any genetic predisposition associated with TMJa. Their study reported 2

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Fig. 1. [A-E]: Characteristic features of paediatric ankylosis - Mandibular micrognathia, facial skeletal and soft tissue asymmetry, malocclusion and reduced mouth opening.

polymorphisms (rs2073618 and rs2228568) and the mutations rs202090603 (p.E33K), rs140782326 (p.V281 M), rs11573942 (p. L295), and rs1375250340 (p.I389T) in the TNFRSF11B gene associated with TMJa. 57

3. Classification of ankylosis

Based on radiographic features as seen using cross sectional imaging (CT & MRI) ankylosis has been classified as follows:^{4–6} Type I/A1.

- Presence of fibrous ankylosis in and around the joint.
- The condyle is flattened and in close approximation to the glenoid fossa; with a reduced joint space.
- The disc is intact in most cases and displaced anteriorly/anteromedially along with the displaced fragment.

Type II/A2.

- Characterized by limited bony bridging between the condylar head and the articular surface along the lateral aspect.
- The condyle is flattened and in close approximation to the glenoid fossa; with a reduced joint space.
- Bifid or enlarged condyle is typical on CT scans.
- The disc is intact in most cases; and displaced anteriorly/anteromedially with displaced fragment.
- The residual condyle fragment, if present, is bigger than 0.5 of the condylar head in the medial side.

Type III/A3.

- Characterised by a bony block bridging across the ramus of the mandible and the zygomatic arch.
- The displaced head is atrophic and lying either free or fused with the medial side of the upper end of the ramus.
- On the deeper aspect, the upper articular surface and disc are intact.
- The residual condyle fragment, if present, is smaller than 0.5 of the condylar head in the medial side.

Type IV/A4.

• Characterised by a bony block that is wide and deep and extending between the ramus and upper articular surface; completely replacing the architecture of the joint.

4. Investigations

- 1) RADIOGRAPHIC INVESTIGATIONS:
 - a. **CT** to assess location and dimensions of the ankylosis and other relevant anatomical features [Fig. 2].
 - b. MRI to assess disc status & position.
 c. Scintigraphy: The authors have observed (Unpublished data) that scintigraphy shows increased activity in ankylotic mass in recurrent cases with large opposing surfaces indicative of high osteogenic potential.
 - d. **OPG** for assessment of dental status.
 - e. Lateral Cephalogram for assessment of facial profile & pharyngeal airway space [Fig. 3].
 - f. **CT/MRI angiography** for assessment of vascular structures in close proximity to ankylotic mass.
- 2) BLOOD & SEROLOGICAL INVESTIGATIONS

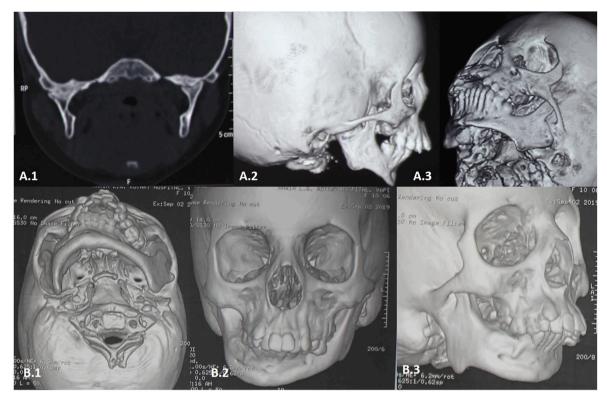


Fig. 2. [A-B]: Characteristic CT scan appearance of paediatric ankylosis – Reduced to absent joint space, skeletal asymmetry, destruction of TMJ architecture, dental malocclusion, elongated coronoid process and bowing of the mandibular corpus [in unilateral ankylosis cases]. Patient A is a case of bilateral ankylosis. Patient B is a case of unilateral ankylosis.

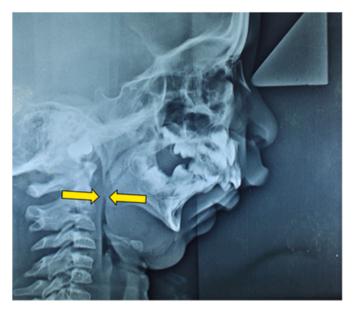


Fig. 3. Lateral cephalogram of a paediatric ankylosis case showing reduced posterior airway space on account of the retruded mandible.

- a. Routine pre-operative blood Investigations
- b. **Serological investigations** to rule out systemic disease in patients with systemic arthropathy

3) POLYSOMNOGRAPHY STUDIES:

For children with history of snoring and day time somnolence, sleep studies are conducted for at least 6 h to assess Apnoea-Hypopnea Index, O₂ Saturation, Snoring, Electroencephalography, Electromyography and Thoracic, Abdominal and Limb Movements.⁷

4) MEDICAL REFERRALS:

To appropriate medical specialities e.g., ENT, Rheumatology, Psychology, Dietician, Physiotherapist, Paediatrician and Anaesthetist etc.

5. Pre-operative considerations

1) INDICATION FOR SURGICAL INTERVENTION:

Mouth opening less than 25 mm for more than 2 months [without significant improvement from jaw opening exercises] can be considered as a baseline indication for surgical intervention.⁴

2) AGE OF RELEASE:

The severity of facial asymmetry, elongation of coronoid process, degree of muscle atrophy, intensity of psychological stress and difficulty of surgical correction are directly proportional to the number of years of ankylosis; especially in paediatric patients. One would assume that early surgical release would provide the best treatment outcome; however, long term success of an optimal surgical outcome is dependent on the patient's compliance to jaw physiotherapy. Patient compliance is correlated to the patient's age, psychological development, underlying systemic and/or physical conditions and support and co-operation from the parents. To satisfactorily meet the above-mentioned conditions, the recommended minimum age for surgical release is 3 years.^{8,9}

The patients treated at the authors centre generally belong to lower socio-economic strata; with working parents unable to supervise daily jaw physiotherapy. The author faced extremely high re-ankylosis rate (100%) for children below the age of 5 years. For this reason, children below 5 years of age were kept under observation, performing TMJa

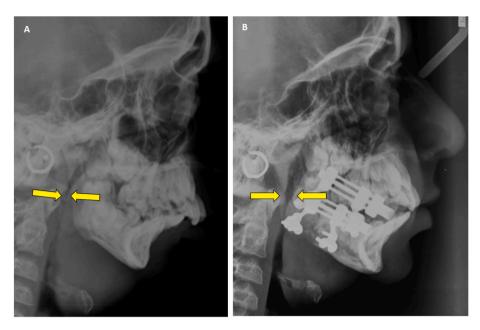


Fig. 4. Distraction Osteogenesis of the mandible to increase the posterior airway space and reduce severity of obstructive sleep apnoea; prior to ankylosis release.

release only when support and psychological development was deemed adequate for the success of the procedure.

5.2. Performing DO AFTER ankylosis release

There are a number of disadvantages associated with this technique:

3) SLEEP APNOEA STATUS:

The shortened, retrusive mandible in TMJa patients causes backward-displacement of the tongue and simultaneous reduction of the upper oropharyngeal airway. This can induce obstructive sleep apnoea syndrome (OSA). Due to the long-term detrimental effects associated with OSA, the priority in TMJa patients with OSA is to relieve this as soon as feasible. Analysis of the pharyngeal airway and polysomnographic studies are mandatory in the management of TMJ ankylosis.

Patients are generally graded using the Paediatric Epsworth Sleep scale & AHI-PAS [Pharyngeal airway space] scale.^{7,10}:

- a. Mild: AHI: 5–15, PAS >8 mm
- b. Moderate: AHI: 15-30, PAS: 5-7
- c. Severe: AHI: >30 PAS: <4 mm Sleep apnoea can be corrected by^{3,7,10–14}:
- a. Continued growth of the mandible (following release of TMJa in paediatric patients)¹¹
- b. Continued Growth by reconstruction of TMJ with a growth centre (Costochondral grafting)
- c. Myofunctional appliance therapy³
- d. Genial advancement¹²
- e. Maxillo-mandibular advancement
- 1) Orthognathic surgery^{13,14}
- 2) Distraction osteogenesis^{7,10}

5.1. Distraction Osteogenesis (DO)

The purpose of distraction at this stage is neither TMJ reconstruction nor cosmetic correction; but relief from OSA. Any cosmetic improvement is a secondary gain. It is important to note that mandibular body DO [horizontal advancement] primarily provides relief from OSA. This is not to be confused with mandibular ramus DO performed for TMJ reconstruction.

There are certain controversies in the application of DO which have been briefly discussed below. $^{7,10}\!$

- i. Poor compliance to post-operative jaw physiotherapy has been observed in TMJa patients who underwent ankylosis release without correction of *pre-operative moderate to severe OSA*. This has been attributed to stimulation of the trigemino-cardiac reflex beyond a certain point of jaw stretching exercises on account of the reduced posterior pharyngeal airway space. Such non-compliance can theoretically lead to early re-ankylosis.
- ii. In cases treated by gap arthroplasty, the lack of a posterior fixed stop makes controlling the DO vector difficult, increasing the risk of a suboptimal result.
- 5.3. Performing DO At The Time of ankylosis release

There are a number of disadvantages associated with this technique:

- i. The presence of a distractor in the tooth bearing segment may interfere with active jaw physiotherapy.
- ii. Need for callous moulding during the active distraction and consolidation phases may interfere with active jaw physiotherapy.
- iii. Need for active jaw physiotherapy may alter the vector of distraction due to the generation of external forces.
- Need for active jaw physiotherapy may damage the callous at the osteotomy site; leading to pseudo-arthrosis.

5.4. Performing DO BEFORE ankylosis release

To reduce the above-mentioned issues, it was proposed that horizontal distraction of the mandible should be performed initially to correct micrognathia and relieve the underlying OSA; followed by ankylosis release. This has numerous advantages:

- i. Provides an opportunity for early relief of OSA.
- ii. The ankylotic mass acts as a stable posterior stop and provides reliable vector for distraction.



Fig. 5. Modified cut for gap arthroplasty to prevent posterior bony contact at the time of maximal mouth opening.

- iii. Early partial correction of facial deformity contributing to improved patient compliance due to alleviation of the associated psychological stress.
- iv. Prevents respiratory embarrassment during physiotherapy and increases compliance to active jaw physiotherapy.
- v. Provides an opportunity to avoid a third surgery for distractor removal; as distractor device removal and ankylosis release is planned simultaneously.

In the author's centre, non-OSAS and mild OSAS patients (PAS>8 mm, AHI 5–15) were treated by direct release of ankylosis followed by active jaw physiotherapy with satisfactory long-term results. DO was performed if the child developed **moderate or severe OSA** at an early age [less than 5 years]. Ankylosis release was subsequently performed after 5–6 years of age; when they would comply better with jaw exercises [Fig. 4].

6. Treatment

- 1. THE AIMS AND OBJECTIVES FOR TREATMENT OF TMJa:
 - i. Increasing the range of mouth opening and improving mandibular function.
 - ii. Decreasing disability and pain; thus, improving quality of life.
 - Preventing recurrence from ectopic bone formation around the joint.
 - iv. Restoring the growth potential of the ramus in growing patients.
 - v. Correction of obstructive sleep apnoea syndrome.
 - vi. Refining of facial aesthetics once growth is complete.
 - vii. Creation of an acceptable and functional dental occlusion.
- 2. CORNERSTONES ON WHICH TREATMENT SHOULD BE BASED:¹⁵
 - i. Minimal intervention.
 - ii. Minimal economic burden.
 - iii. Minimal morbidity.
- 3. GENERALLY ACCEPTED TREATMENT PROTOCOL:

The generally accepted protocol for achieving maximal mouth opening intraoperatively is as follows^{3,8,16-18}:

- i. Essential Wide and aggressive gap creation of 1-1.5 cm.
- ii. Essential Ipsilateral coronoidectomy to further improve mouth opening to around 35 mm; especially in cases with an elongated coronoid process. Contra-lateral coronoidectomy; if steps 1 and 2 do not result in a mouth opening of 35 mm.
- iii. Preferred Interposition of a suitable biological material to reduce dead space and prevent recurrence. The native articular disc is most preferred; if present and salvageable.
- iv. Optional Stripping of the pterygo-masseteric sling [preferred in long standing cases].
- v. Optional Reconstruction of the TMJ post excision of the ankylotic mass:
- a) Primary TMJ reconstruction of the condyle ramus unit with autogenous grafts or distraction osteogenesis is commonly performed.
- b) Secondary TMJ reconstruction on completion of facial growth [after 18 years of age] is an accepted alternative line of treatment as well.
- c) It is important to note that total joint replacement with an alloplastic prosthesis is currently recommended only for adult patients.

Roychoudhury et al. 2014¹⁹ has suggested certain modifications to the routine surgical technique while creating the bony gap to help reduce the risk of re-ankylosis:

A piezoelectric osteotome may be used over conventional rotary cutting tools as it -

- a) Causes minimal bleeding.
- b) Minimises damage to adjacent soft tissues and vital structures.
- c) Minimises undesirable seeding of cortical bone chips into the surrounding area.
- d) Improves manual dexterity when working in an anatomically delicate area.

- e) Leaves smooth bone ends after cutting.
- f) Allows for safe removal of bone from the medial aspect.

This leads to a reduction in overall post-operative complications. The disadvantages associated with this device is the increased intraoperative time and initial monetary investment.¹⁹

When a gap is made by making parallel osteotomy cuts to excise the ankylotic mass, a bony contact may occur between the posterior aspect of the upper and lower osteotomy cuts on maximal mouth opening [Fig. 5]. Thus, the lower osteotomy cut is modified by making the posterior one-third cut divergent. This eliminates the bony contact during maximum mouth opening and may theoretically reduce the chances of re-ankylosis.²⁰

4. TREATMENT OPTIONS CAN BE ARTIFICIALLY DIVIDED INTO:

- i. Resective Procedures:
 - A. Minimal Arthroplasty (MAP)
 - B. Lateral Arthroplasty (LAP)
 - C. Gap Arthroplasty (GAP)
 - D. Sub-ankylotic Arthroplasty (SAP)
- ii. Reconstructive Procedures:
 - A. Interpositional material
 - B. Autogenous Reconstruction
 - C. Alloplastic Reconstruction
- I. Resective Procedures:
 - A. Minimal Arthroplasty (MAP)

Minimal Arthroplasty is mainly indicated for fibrous ankylosis and includes procedures like brisement force mouth opening under general anaesthesia, arthroscopic adhesion lysis, high condylar shave and fibrous tissue excision and re-contouring of the articular surface.

B. Lateral Arthroplasty (LAP)

The LAP technique was first proposed by Nitzan. Total resection of the ankylotic bridge laterally around the residual condyle and interposition of the residual articular disc are the prime determinants for successful treatment. Type 2/A2 cases are ideal candidates for LAP. Mild forms of Type 3/A3 may also be candidates for LAP.^{4–6} Improvement in facial symmetry and growth potential have been reported in growing patients after performing LAP.^{9,21–25}

Advantages:9,21-25

- 1. Condylar function preserved.
- 2. Growth potential of the residual condyle is preserved.
- 3. No need of joint reconstruction.
- 4. Shorter Surgery time.
- 5. Less risk of injury to medial structures.
- 6. No/minimal loss of ramal height.
- 7. Occlusion is maintained.
- 8. Less incidence of re-ankylosis.

Disadvantages:^{9,21–25}

- 1. Difficult to visualise residual condyle
- 2. Damage to residual condyle
 - C. Gap Arthroplasty (GAP)

The block of bone between the skull base & the ramus is completely excised to create a functional gap between the joint components. Type 3/A3 and Type 4/A4 are ideal candidates for this technique.^{4–6} [Fig. 5]

The greater the distance between the cut surfaces of the ramus and the skull base, the less the risk of re-ankylosis. However, excessive excision of bone would lead to a loss of ramal height and lack of support for the rotating mandible, presenting as contralateral open bite and deviation on mouth opening. Thus, the amount of bone removed is crucial to the success of treatment.²⁶ Studies have documented a minimum recommended gap to prevent re-ankylosis.

- 1. 5–9 mm^{16,26,27}
- 2. 10–14 mm^{28–30}
- 3. 15–20 mm^{3,8,31}

The primary difficulties noted in performing a successful GAP is the abnormal and unpredictable anatomy of the joint and proximity to vital structures along the medial aspect of the joint. Any residual bone left along the medial aspect will make post-operative physiotherapy painful, which can lead to non-compliance and early re-ankylosis. The presence of any residual bone in the gap can increase the risk of re-ankylosis.^{8,32,33}

Advantages:^{3,8,11,29,34–36}

- 1. Possibility of neo-condyle formation.
- 2. Simple surgical procedure.
- 3. Short operating time.
- 4. Less blood loss.

Disadvantages:^{3,8,11,29,34–36}

- 1. Radical surgical removal.
- 2. Risk of injury to medial structures.
- 3. Further shortening of ramus.
- 4. Worsening of malocclusion.
- 5. Pit defect in the back of the jaw.
- 6. Higher rate of re-ankylosis.
- 7. Requirement of extensive jaw physiotherapy.
- 8. Risk of leaving bone along the medial aspect of the joint D. Sub-Ankylotic Arthroplasty (SAP):

In supero-inferiorly and medio-laterally large ankylotic masses (Sawhney's IV; especially with a history of re-ankylosis) an aggressive GAP entails the following:^{4–6}

- 1. Large extent of removal that poses considerable risk to vital structures; increasing the risk of intra-operative and post-operative complications.
- 2. Large apposing raw bone surfaces that heal by scarring.
- 3. Interpositional material rarely able to bridge the large area.
- Excessive loss of ramal height; leading to loss of mandibular stability and malocclusions.

These kinds of post-operative conditions can theoretically lead to an increased risk of re-ankylosis. Thus, the concept of sub-ankylotic arthroplasty came about. In SAP, the gap is created below the ankylotic mass. The recommended gap at the osteotomy site is 5–6 mm. Alternatively, one can achieve a gap without removing bone as well; by stripping the pterygomasseteric sling and distracting the mandible downwards to create space. A passive mouth opening of at least 3 cm can be achieved through SAP.^{26,37}

Advantages:^{26,3}

- 1. Surgical approach is easier.
- 2. Less risk of injury to medial structures.
- 3. Smaller raw opposing surfaces.
- 4. Lower risk of re-ankylosis.
- 5. Less operating time.
- 6. Gap need to be made compulsorily.
- Ipsilateral coronoidectomy may not be required. Disadvantages:^{26,37}
- 1. Original pathology cannot be assessed as it is not excised.
- 2. Location of joint inferior to contralateral side; long term consequences unknown at this point.

In the author's centre, a large ankylotic mass exhibiting an increased uptake on scintigraphy with a history of recurrence is an ideal candidate for SAP.

II. Reconstructive Procedures:

Given the extensive nature of TMJ reconstruction, the current review will be restricted to the management of paediatric ankylosis alone. An attempt will be made to review the most commonly used interpositional materials and also the rationale behind inter-positional arthroplasty. TMJ reconstruction using autogenous vascularised and non-vascularised grafts, distraction osteogenesis and total joint replacement techniques will be covered elsewhere in detail in the current issue.

6.1. Interpositional materials

There are various interpositional materials:³⁸

- 1) No Interpositional material (essentially plain gap arthroplasty)
- 2) Autogenous materials
 - a. Cartilage [Ear/Rib/Sternum]
 - b. Fascial/Myofascial/Muscle flap [Temporalis/Masseter/Fascialata]
 - c. Fat graft [Plain Fat/Dermis-fat/Buccal fat pad]
 - d. Skin graft [Full thickness/Dermis]
- 3) Alloplastic materials
 - a. Silastic
 - b. Teflon-proplast
 - c. Methyl methacrylate
 - d. Fossa prosthesis
- Allogenic materials [Lyophilized, Freeze-dried, Cryo-preserved]
 a. Cartilage
 - b. Dura
- 5) Xenograft materials [Bovine/Porcine in origin]
 - a. Cartilage
 - b. Collagen
- 6.2. Main criteria for choosing the material $are^{29,38}$
 - 1. Easy graft harvesting technique.
 - 2. Cosmetic consequences of graft harvesting.
 - 3. Donor site morbidity.
 - 4. Abundantly available.
 - 5. Adequate bulk.
 - 6. Good handling properties.
 - 7. Low risk of infection.
 - 8. Durability.
 - 9. Tolerance.
 - 10. Biocompatibility.
 - 11. Cost of material.
 - 12. Facilitates normal joint function.
 - 13. Protects condyle from severe remodelling.
 - 14. Prevention of recurrence.

6.3. Common interpositional materials

i. Rationale Behind Interpositional Arthroplasty:

Extensive resection of the ankylotic mass leads to the presence of considerable dead space and large raw bony surfaces. If no effort is made to fill up the dead space or line the raw surfaces, hematoma formation with subsequent organisation takes place. Local pluripotent stem cells may then be induced to differentiate into fibroblasts and osteoblasts, with deposition of collagen and bone. In excessively fibrotic joints, there is a decrease in vascularity; and thereby, a decrease in oxygen tension in the surrounding tissues. Decreased vascularity and oxygen tension in the

surrounding tissues may additionally favour the transformation of fibrous tissue into cartilage and bone. $^{39}\,$

The presence of an interpositional material theoretically reduces the risk of re-ankylosis by reducing the dead space, lining the raw cut bony surfaces and reducing the volume of hematoma formation. Materials that are anti-osteogenic [e.g., dermis fat] should serve even better in this regard. 38,40

Matsuura et al., 2001 studied the functional and anatomic changes after GAP in TMJa animal models and showed that GAP was unable to restore the TMJ to its functionally and histologically pre-existing state.³⁶

A systematic review and meta-analysis of the clinical outcomes for various surgical modalities in the management of TMJa by Al-Moraissi et al., 2015 showed that interpositional arthroplasty was superior to plain gap arthroplasty with respect to maximal mouth opening and recurrence rate with the difference being statistically significant.⁴¹ Topazian et al., 1966 echoed similar findings with an unacceptably high recurrence rate of 53% in the gap arthroplasty group compared to 0% in the interposition group.³⁵ Roychoudhury et al., 2021 On the other hand, Roychoudhury et al. 2014,⁴² Andrade et al. 2020³⁰ and Roychoudhury et al. 2021⁵⁰ documented that interpositional arthroplasty with temporalis myofascial flap, abdominal dermis fat and costochondral graft respectively were not statistically superior to plain gap arthroplasty with respect to recurrence rates and maximal mouth opening.

Thus, there is considerable debate regarding the efficacy of interpositional and plain gap arthroplasty in the literature.

ii. Temporalis Myofascial Flap (TMF):

The TMF is the most commonly used interpositional material today. The flap is pedicled inferiorly on the medial temporal artery. Once an adequate length of the flap is mobilised, it is rotated below the zygomatic arch and into the gap, where it is secured to the surrounding soft tissues. The bulk of the graft required depends on the volume of the defect. At the least, the deep temporalis fascia and the superficial muscle layer are harvested to support joint function and to maintain flap vascularity.^{27,41,43,44}

Advantages:^{26,27,40–44}

- 1) Close proximity to the site.
- 2) Good and dependable vascular supply.
- 3) Documented long term viability.
- 4) Ease of access as same incision is used.
- 5) Easy preparation of graft.
- 6) Adequate bulk of graft available.
- 7) Minimal risk of nerve damage.
- 8) Good functional and clinical results.
- 9) Minimal complications.

Disadvantages:^{26,27,40–44}

- 1) Cosmetic consequences of temporal hollowing.
- 2) Temporalis muscle atrophies to fibrous tissue in the long run
- Fibrosis and scar contracture of the temporalis muscle, which may cause trismus.
- 4) Chronic headache in the long term.
- 5) Tissues may be too soft to resist the compression of the ramus.
- 6) Open bite due to loss of elevator action by harvesting temporalis.
- 7) Increase risk of damage to facial nerve branches due to superior extension of the pre-auricular incision in the temporal region along with greater exposure needed.
- iii. Abdominal Dermis-Fat graft:

Dermis fat graft harvested from the abdomen is gaining popularity as an interpositional material for TMJa. The presence of the 'dermis' serves multiple functions. When attached to the dermis, fat tends to be more stable and less likely to break apart while handling. The dermal layer



Fig. 6. Abdominal Dermis Fat Graft Harvest and Interposition into the gap. Adequate bulk of graft is seen; and is passively filling up the entire joint space.

also serves as the vaso-inductive layer for the underlying adipose graft. 38,40 [Fig. 6]

Multiple histologic and radiographic studies have documented that the dermis fat graft undergoes an initial period of necrosis and volume shrinkage [up to 33%]. However, in the long term the volume is maintained or even increased [up to 33%] due to neo-adipogenesis. In comparison to dermis fat grafts, studies using plain abdominal fat have been associated with higher shrinkage rates in the long term.⁴⁵

Neo-adipogenesis has an additional benefit of inhibiting the formation of new cartilage and bone within the joint, reducing the risk of reankylosis. The intrinsic pro-coagulant and anti-inflammatory effect also contribute to preventing heterotrophic bone formation. Numerous studies have documented the long-term safety of dermis fat graft for TMJ surgery. In light of this evidence, the dermis fat graft serves as a near optimal interpositional graft material for TMJ surgery.^{38,40,41,45,46}

Advantages: 38,40,41,45,46

- 1) Easily sculpted and trimmed to fit any gap.
- 2) Minimal donor site morbidity as primary closure is possible.
- 3) It does not atrophy with time; and may increase in volume.
- 4) Vascularity does not play an important role as necrosis is part of the neo-adipogenesis process.
- 5) Anti-osteogenic properties that may help reduce the risk of reankylosis: anti-inflammatory action, pro-coagulant action and neo-adipogenesis activity.
- 6) Improved quality of life due to smooth, pain-free function.
- 7) Develops mild fibrotic changes in the long term that help it better cope with the mechanical function of the joint.
- 8) Available in adequate bulk to fill the entire surgical defect [even in large gap arthroplasty cases] and reduce dead space.
- 9) Intrinsic thromboplastin-mediated pro-coagulant activity that helps achieve faster haemostasis and reduce volume of blood clot formation.
- 10) Isolates the adjacent tissues from the joint space, preventing the migration of pluripotent stem cells from the reactive tissues into the joint space to form heterotopic bone.
- 11) Localised anti-inflammatory effect that helps reduce heterotopic bone formation.
- 12) The process of neo-adipogenesis inhibits the formation of new cartilage and bone within the joint.
- 13) Abdominal fat contains large fat lobules that are relatively resistant to resorption.

- 14) Abdominal fat contains more stem cells; which have excellent potential for lipogenesis; this is extremely beneficial in preventing adipolysis in the long term.
- 15) Contour deformities commonly found on harvesting TMF are avoided.
- 16) Decreased risk of damage to facial nerve branches due to minimal extension of the pre-auricular incision in the temporal region along with lesser exposure needed.

Disadvantages:^{38,40,41,45,46}

- 1) Second distant donor site required.
- 2) Cosmetic consequences of the scar on the abdomen.
- 3) Thin patient may not have as much bulk of adipose tissue to harvest requiring a longer incision.
- 4) Risk of idiopathic condylar resorption documented; which may be due to intrinsic factors and not necessarily due to dermis fat.iv. Pedicled Buccal Fat Pad:

Pedicled buccal fat pads are used in oral surgery for reconstruction of a variety of defects. However, their use as an interposition material after gap arthroplasty in cases of ankylosis of the temporomandibular joint (TMJ) is less documented. $^{45,51-55}$

It has been documented that the buccal fat pad mean volume is approximately 10 ml and weighs 9.3 g with little variation in terms of size or weight. It appears to be constant among different persons and it is described even in cachectic patients with poor subcutaneous fat. It is capable of covering defects of about 4 cm in diameter.⁵⁵ Advantages:^{45,51–55}

- 1) Proximity to the surgical defect.
- 2) Pedicled blood supply.
- 3) Easy availability.
- 4) Decreased risk of damage to facial nerve branches due to minimal extension of the pre-auricular incision in the temporal region along with lesser exposure needed.

Disadvantages: 45,51–55

- Volume of buccal fat pad is less and hence can only be utilised in mild ankylosis cases where minimal gap arthroplasty defects [5–7 mm] will be created with a defect volume of less than 7 ml.
- The exact volume of the buccal fat pad cannot be measured intraoperatively because it is a pedicled graft.

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Mouth opening after 3 years

Mouth opening after 2 years Mo

Mouth opening after 7 years

Mouth opening after 3 years

Fig. 7. Long term follow up of paediatric TMJ Ankylosis cases.

- 3) Greater amount of shrinkage [approx. 67.5%] and lower retention rate [approx. 32.44%] has been documented in the long term.
- 4) The buccal fat pad should be evaluated pre-operatively to ensure that sufficient volume will be available during surgery.
- 5) Controversy exists regarding actual volumes of buccal fat pad being found in TMJ ankylosis patients on account of being on a poor nutritional diet for long periods.⁵³
- 6) Contour deformity following the removal of buccal fat pad could be a cause for concern.

7. Post-operative care

Long term active jaw physiotherapy is most important to maintain an optimal surgical outcome. Longstanding ankylosis causes a lack of tonus in the closing muscles, because the mandible was supported by the ankylosis. Jaw physiotherapy also enhances muscle bulk and strength and improves range of motion following a prolonged period of relative inactivity of the surrounding mandibular musculature.⁴⁰

Physiotherapy should be started as early as possible in order to disrupt and prevent adhesions, prevent soft-tissue contractions and redevelop normal muscle function. While some choose to start physiotherapy in the immediate post-operative phase [same day to within 48 h of surgery]; others may delay for up to a week. This rest period allows pain and swelling to reduce, increasing patient compliance. It also reduces the risk of reactionary bleeding and allows for early phase healing of the surrounding tissues and the interpositional graft material. Initial exercises are of a passive nature and include hinge opening and lateral excursions combined with manual finger stretching in front of a mirror. These should progress to aggressive jaw physiotherapy after a few weeks. Aggressive jaw physiotherapy involves jaw exercises of greater intensity and frequency; and may be supported by a Heister jaw stretcher or a Thera-Bite Jaw Rehabilitation System (Craniomandibular Rehab, Denver, CO).^{3,8,18,30,40}

Mouth opening after 3 years

At the author's centre, passive jaw physiotherapy is started on the first post-operative day. Aggressive jaw physiotherapy is started from the fifth post-operative day for a period of 3–4 weeks until a passive pain-free mouth opening of at least 30 mm is achieved.³⁰ [Fig. 7]

Some TMJ reconstruction modalities require a period of immobilization before starting jaw physiotherapy. Costochondral grafting requires a period of maxilla-mandibular fixation for up to 10 days. In

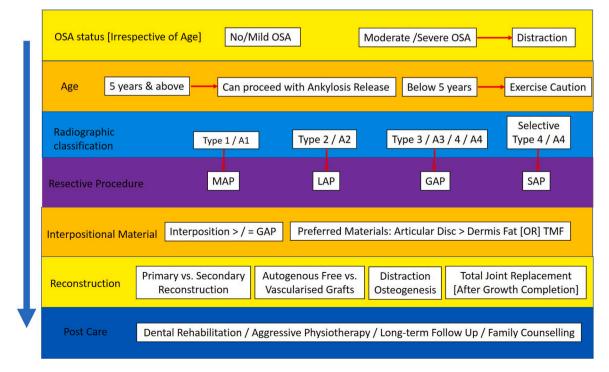


Fig. 8. Flow Chart summarizing treatment protocol for Paediatric TMJ Ankylosis [TMF = temporalis myofascial flap, GAP = gap arthroplasty, OSA = obstructive sleep apnoea].

comparison, TMJ reconstruction with DO permits immediate post-operative physiotherapy. $^{\rm 8}$

Failure to achieve the documented intraoperative maximal mouth opening, or if the maximal mouth opening shows no sign of improvement at 6–8 weeks, the jaw should be stretched with the patient under general anaesthesia.⁸

For the next 6–12 months the patient should continue jaw physiotherapy throughout the day; at a similar intensity but a reduced frequency. The physical therapy program may also include heat, massage, and gum chewing. Ultrasonic wave or infrared ray therapy with calcium or iodide infiltration deep into the joint may be used as adjuvants during physiotherapy.

After release of longstanding ankylosis changes may occur in the occlusion; which are more pronounced following bilateral arthroplasties. These situations are solved by guided physiotherapy and myofunctional orthodontic therapy. The patient also needs to be trained to bite into an acceptable occlusion. Split therapy may be initiated in the post-operative phase and should remain in place for three months, without adjustment, to maintain an open bite. The splint is then gradually adjusted by grinding off the maxillary side to allow eruption of the maxillary teeth and to close any residual open bite.^{4,8,40}

Undiagnosed depression can be a significant obstacle in the patient's rehabilitation following ankylosis release. Thus, sessions of psychological counselling for the patient and family members generate an optimal environment for recovery.⁴⁰

8. Conclusion

TMJ ankylosis is a crippling craniofacial deformity; most often presenting as complication of paediatric condylar trauma. Evaluation and diagnosis are essential for appropriate management. Special attention should be paid to OSA status and the age of intervention. Evidence-based management mandates selection of appropriate resective and reconstructive techniques. The current review of the literature, though divided, reveals a preference for interpositional arthroplasty over plain gap arthroplasty; with respect to long term results related to maximal mouth opening and risk for re-ankylosis.^{41,47–49} It is prudent to note that aggressive jaw physiotherapy in the postoperative phase is of utmost importance to maintain the results of an optimal surgical outcome [Fig. 8].

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Declaration of competing interest

None.

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- 56 Paediatric Temporomandibular Articulation Ankylosis: OPG and RANKL Gene Expression Analysis. André da Silveira Braune1, Patrícia Cataldo de Felipe Cordeiro2, Letícia Ladeira Bonato3, Ricardo Lopes Cruz1, Jamila Perini1, Valquiria Quinelato2*, Jose de Albuquerque Calasans-Maia2 and Marco Bernardo Cury Fernandes 7 - Issue 5 DOI: 10.31031/OPROJ.2020.07.000671, Orthopedic (Research Online Journal).
- 57 Mutations in the osteoprotegerin-encoding gene are associated with temporomandibular joint ankylosis. Paola FernandaCorsoDDS, MSc, PhDa Renato Assis Machado DDS, MSc, PhDbc JenniferTsi Gerber DDS, MSca Tatiana Miranda Deliberado rDDS, MSc, PhDdDavani https://doi.org/10.1016/j.oooo.2021.08.017.