Management of Young Patients with Temporomandibular Joint Ankylosis—a Surgical and Anesthetic Challenge

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Abstract Temporomandibular joint ankylosis is a unique disease where fracture of the mandibular condyle or any other cause leading to ankylosis of the joint can lead to multiple problems if not detected and treated early. If affected in early years of life, it may cause facial dysmorphism, restricted mouth opening, and difficulty in eating, speech, and sleep. Early surgery and physiotherapy can restore the joint function to a great extent. Anesthetizing a pediatric patient with this disorder is a definite challenge which needs expertise in difficult airway management.

Keywords Difficult airway · Young patients · Anesthesia in TMJ ankylosis · Surgery in TMJ ankylosis

Introduction

Trauma is the major cause of temporomandibular joint (TMJ) ankylosis globally [1–4]. Post traumatic TMJ ankylosis is on rise especially in developing countries like India, due to the delay in treatment as these closed condylar fractures are not easily apparent [1]. Also in developing nations as access to specialized healthcare from remote areas is difficult, often, parents seek an opinion only when the problem is at an advanced stage and mouth opening is almost negligible [5, 6].

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Pathophysiology of TMJ Ankylosis

Fracture of the mandibular condyle in childhood and subsequent organization of the hematoma result in formation of varying sizes of juxta-articular bone and ankylosis, or consolidation and immobility of the joint. The growth of bone in juxta-articular tissue may be similar to that seen in myositis ossificans traumatica, often seen following traumatic elbow fractures wherein an osseous bridging is formed without any effect on the anatomical condition of the joint [7].

Though trauma is the most frequent cause of TMJ ankylosis (13–100 %) [8], incidence attributed to different causes are local or systemic infection (10–49 %), or systemic autoimmune diseases (10 %) such as ankylosing spondylitis, rheumatoid arthritis, psoriasis, etc. Previous surgery on TM joint can also lead to reankylosis [9, 10]. Other causes may be congenital malformations, use of forceps for delivery (2.6 %) [6], infection (e.g., ear suppuration) [11, 12], rarely neoplastic [13], or unknown etiology.

Clinical Presentation

The usual age of onset of TMJ ankylosis in children is below 10 years [14] concurrent with the active growth phase of early childhood [10]. Children present with restricted mouth opening; dentofacial deformity; malocclusion; poor oral hygiene; dental caries; cosmetic disability; impaired speech; and difficulty in mastication, malnutrition, and obstructive sleep apnea [9, 13]. Patients with bilateral TMJ ankylosis often have micrognathia and retrognathia and have symptoms of severe obstructive sleep apnea (OSA) [15]. Classically, patients with chronic long-standing TMJ ankylosis have been described as having “bird-faced” deformity with convex facial profiles, characterized by small and receding mandible with steep occlusal plane. The maxilla, soft tissues enveloping the mandible

and suprahyoid muscle is all secondarily affected. The oropharyngeal airway is narrowed secondary to shortening of the mandibular rami with narrowing of the space between the mandibular angles [16] (Fig. 1). A clinical indicator of the severity of ankylosis is the interincisal opening; when the opening is less than 5 mm it is termed as a “complete ankylosis” [10, 12].

Radiological Diagnosis

Computed tomography (CT) scans consisting of coronal sections (Fig. 2) of the TMJ along with and three-dimensional views (Fig. 3) are considered essential for preoperative radiographic evaluation as well as postoperative assessment of the surgical procedure. Orthopantomogram (Fig. 4) is an inexpensive and important diagnostic aid. It is a panoramic radiograph of maxilla and mandible in two dimensions. It gives a good assessment of the coronoid process and comparison with the coronoid process of the contralateral side. It also helps to study the dental component and associated structures.

Surgical Techniques

The treatment of TMJ ankylosis in children is much more challenging than in adults because of high chances of recurrence and unpredictable growth of the mandible. Surgical treatment of ankylosis in children has three basic objectives: the primary objective is to release the ankylosis, second to improve mandibular function, and last to maintain normal growth and development of the face. Children who develop ankylosis before the age of 5 are the most susceptible to facial deformities. Early surgical intervention can reduce the adverse effects on facial development.

There have been different schools of thought with regard to whether the condylar cartilage is a primary growth center and whether its removal would affect the subsequent growth of the mandible. According to the functional matrix theory given

Fig. 1 TMJ ankylosis patient with nil mouth opening

Fig. 2 CT scan showing (coronal section) of TMJ ankylosis patient showing ankylotic bony mass in Bilateral TMJ ankylosis patient

by Moss and Salantijn, growth and maintenance of skeletal tissues are always secondary, compensatory and mechanically obligatory responses to prior events and processes occurring in related non-skeletal tissues, organs, and functioning spaces. He claims that growth of the skeletal components whether endochondral or intramembranous in origin, is largely dependent on the growth of functional matrices. Hence, he theorized that growth of the face occurs as a response to functional needs and is mediated by soft tissues in which the jaws are embedded [1, 17].

A variety of surgical flaps have been described for exposing the TMJ via a pre-auricular approach with temporal extensions. Sufficient exposure requires a flap that can be extended to the size and extension of the ankylotic mass. The classic “question-mark” incision fulfills this requirement [18].

The size and extensions of the bony mass governs the extent of bony resections. The treatment options vary according

Fig. 3 CT scan 3D reconstruction of TMJ ankylosis patient showing bony union between the mandible and base of skull

Fig. 4 Orthopantomogram showing radio-opaque mass in bilateral TMJ region

to complexity of the ankylosis. TMJ ankylosis treatment reflects the complexity of the problem. Treatment options that have been described are gaparthroplasty, interpositional arthroplasty, and excision of the ankylotic mass. Reconstruction of the ramus/condyle unit with autogenous bone, such as a costochondral graft (CCG), fibula, clavicle, iliac crest, metatarsal head, or alloplastic material has all been reported. The clinical and advanced radiographic imaging technique CT scans provides relevant information about the location and extent of the ankylotic bony mass. However, no single method has produced uniformly successful results.

Sawhney described four types of TMJ ankylosis and recommended selection of the appropriate surgical treatment based on the type of defect. Type I: Presence of fibroadhesions at the condyle; Type II: Bone fusion with condyle remodeling and an intact medial pole; Type III: Ankylotic mass, mandibular ramus union with the zygomatic arch and medial pole intact; Type IV: Complete ankylotic mass, total union of the mandibular ramus with the zygomatic arch.

In the treatment of ankylosis types I and II, recontouring, high-condylar shave, and gap arthroplasty have been recommended in children. The surgical treatment of type III and IV ankylosis includes removal of sufficient bone to create a gap and placement of an interpositional material to prevent reankylosis and minimize loss of the posterior dimension of the ramus [19].

Kaban et al. gave extensive and generalized protocol for the management TMJ ankylosis in children. The 7-step protocol is as follows: (1) aggressive excision of the fibrous and/or bony ankylotic mass; (2) coronoidectomy on the affected side; (3) coronoidectomy on the contralateral side, if steps 1 and 2 do not result in a maximal incisal opening of more than 35 mm or do not result in dislocation of unaffected contralateral TM (once the contralateral joint starts dislocating, further increase in mouth opening cannot be achieved; hence, that incisal opening will be considered maximal even if it is not within

the ideal range. (4) Lining of the joint with a temporalis myofascial flap or the native disk, if it can be salvaged. (5) Reconstruction of the ramus condyle unit with either distraction osteogenesis or a CCG and rigid fixation; (6) early mobilization of the jaw; and (7) aggressive physiotherapy [20, 21].

The treatment of ankylosis in children is further complicated by the lack of compliance for physiotherapy. Patient's parents have to be counseled enough prior to surgical treatment. Children 3 years of age and older can cooperate with physiotherapy, provided the ankylosis release is successful and excessive force is not needed to mobilize the jaw during the postoperative period. (Figs. 5, 6).

Complications

The incidence of facial nerve injuries or one of its branches according to various reports ranges from 1 % to as high as 55 % for TMJ surgery. Failure to fulfill the protocols for TMJ ankylosis surgery might result in serious facial nerve injury, which could be resolved only by means of microsurgical techniques for repairing the damaged nerve [22]. However, most injuries produce only temporary motor dysfunction caused due to retraction injury intra-operatively that resolves within 6 months.

Anesthetic Implications

Anesthesia in pediatric patients with TMJ ankylosis is a definite challenge. Technically, it encompasses both the management of a pediatric patient and a difficult airway scenario. Physiological and anatomical differences exist between the adult and the pediatric patient and these differences have to be borne in mind while considering pediatric patients for anesthesia.

Fig. 5 TMJ ankylosis patient after gap arthroplasty with adequate mouth opening

Fig. 6 Orthopantomogram after gap arthroplasty showing gap between base of skull and ramus of Mandible in bilateral TMJ region

Airway Management

As such, in younger children (especially infants), the larynx is cephalad in position as compared to an adult, the epiglottis is large and floppy, and the tongue is larger in relation to the oral cavity. In children less than 6 years, uncuffed endotracheal tubes are preferred because of the funnel-like shape of the larynx. Adenotonsillar hypertrophy is common in preschool children, sometimes making the airway narrowest at the tonsillar level [23]. All these factors lead to a difficult intubation in children.

In addition, the physiological concerns in younger children are of such as higher metabolic oxygen consumption and low functional residual capacity leading to low oxygen reserves. Therefore, there is significant reduction in the time available for performing tracheal intubation and securing the airway in small children [23, 24]. The difficulty in intubation gets further compounded in a child with TMJ ankylosis because of trismus and uncooperativeness [6, 16].

These children tend to have neurobehavioral problems because of the inability to enjoy a normal diet, difficulty in normal speech, sleeping disorders, and facial deformity [13].

TMJ ankylosis patients have increased propensity for airway obstruction and obstructive sleep apnea (OSA) is often associated. This airway obstruction is multifactorial caused mainly due to decreased space in the oral cavity because of encroachment of the oropharyngeal and hypopharyngeal lumen by the tongue and soft tissues of pharynx [16]. OSA in children can be associated with significant morbidity. It is a sleep-related breathing disorder characterized by partial or complete occlusion of the airway during sleep in spite of respiratory efforts. It leads to disparity in gas exchange resulting in hypoxemia, hypercapnia, sleep disturbances, and consequently, daytime somnolence and behavioral issues [25]. Thus, the history of OSA needs to be elicited during the pre-operative assessment to ensure proper intraoperative and post-operative care. Although the most common cause of OSA in children is adenotonsillar hypertrophy, craniofacial

malformations, hypotonia, obesity, midface hypoplasia, macroglossia, retrognathia, micrognathia, glossoptosis, and family genetics are also the accepted risk factor [26]. However, more than one condition can co-exist in pediatric patients with overlapping contributions from adenotonsillar hypertrophy, abnormal airway muscle tone, and genetics [27].

Limited data is available to the real incidence of difficult airway management in pediatric patients [28]. The incidence of difficult tracheal intubation was derived to be around 1.2 to 9 % in pediatric population [29, 30].

Difficult face mask ventilation may also be present. Due to limited mouth opening, insertion of direct laryngoscope may be difficult. Even if that is possible, vocal cord visualization may be difficult or impossible by conventional laryngoscopy. The associated retrognathia with associated pseudomacroglossia narrows down the pharyngeal passage thus further making the visualization of the vocal cords difficult. The various options are available for securing the airway which includes fiberoptic intubation, semi-blind nasal intubation, retrograde endotracheal intubation, bi-nasopharyngeal airway, a fluoroscope-aided retrograde placement of the guide wire for the tracheal intubation, or else a tracheostomy [31].

Review of Methods Used for Securing Airway

Each procedure has some pitfalls. A blind nasal intubation mostly requires repeated attempts using an endotracheal tube introduced through the nares and guided by external palpation of the glottis by the contralateral hand and presence of the end-tidal carbon dioxide trace. As there can be misses, repeated attempts may lead to bleeding and laryngeal edema and loss of the airway in a pediatric patient.

If nasotracheal fiberoptic intubation is planned, care has to be taken to avoid epistaxis. The incidence of epistaxis has been reported to be in the range of 19–54 % with nasotracheal intubation. This bleeding makes it difficult to visualize any structures with the vision of fiberoptic bronchoscope (FOB) making intubation difficult. An awake patient is able to protect against aspiration of blood but this protective reflex is lost once the patient is anesthetized [32].

A retrograde intubation is difficult to perform and hardly applied to a young child with restricted mouth opening. Tracheostomy is mostly reserved for emergency cases as it is an invasive procedure with associated morbidity.

An awake fiberoptic intubation is a gold standard for securing a difficult airway [33]. However, in a pediatric patient, awake fiberoptic intubation is hardly ever possible, and instead, deep sedation or general anesthesia may be needed [24]. With the induction of anesthesia and muscle relaxation, there is a propensity for airway collapse, and in a patient with TMJ ankylosis, there is no scope for jaw thrust and chin lift. The safest option of securing the airway probably is fiberoptic intubation under sedation with spontaneous ventilation.

There are no predictive scores for evaluating the airway in a pediatric patient with TMJ ankylosis. Certain predictors of difficult airway are obvious such as facial dysmorphism, limited mouth opening, restricted mobility of the TMJ, and limited sub mandibular space because of retrognathia and mandibular hypoplasia. Rather, it is an anticipated difficult airway with little or nil mouth opening [28, 34]. Although literature does mention protocols for airway management in syndromic children, there is no mention of children with TMJ ankylosis [23, 35]. Management requires sufficient expertise in handling difficult airway in the pediatric population. It requires sufficient skill to use the various equipments available for managing airway including the FOB. Both skill and a pediatric fibreoptic may be needed for securing the airway in children with TMJ ankylosis. However, anesthetists have devised new techniques of securing the airway even in the absence of appropriate equipment and have developed new methods. We review a few reports.

Due to non-availability of a pediatric FOB, Mohan K et al. used total intravenous anesthesia (TIVA) with fentanyl and propofol in a 12-year-old boy with unilateral TMJ ankylosis planned for condylectomy and interposition arthroplasty. Spontaneous respiration was maintained and oxygen administered via nasal prongs. An extra oral condylectomy was performed under TIVA. Spontaneous respiration was maintained with nasal prongs. Once the mouth could be opened, endotracheal intubation was performed with direct laryngoscopy and gum elastic bougie [31]. But this method may not be appropriate for a child OSA.

Kang JM et al. have described the management of a 34-year-old patient with ankylosing spondylitis and TMJ ankylosis. Intubation was carried out by an anesthetist with an experience of more than 200 awake fibreoptic intubations in order to avoid tracheostomy. An awake fibreoptic intubation was carried out with topical local anesthetics [32]. But the same is not possible in a pediatric patient, who would require some sedation.

In a study by Gupta et al. dexmedetomidine was used for providing conscious sedation for nasotracheal fibreoptic intubation in adult patients with TMJ ankylosis. The dexmedetomidine group was found to have better intubating conditions, more hemodynamic stability, and less adverse effects as compared to the propofol group. The US FDA approval for the use dexmedetomidine is only for sedation in adults undergoing mechanical ventilation for <24 h. However, it is used as an off-label adjunctive agent for sedation and analgesia in pediatric patients in the critical care unit and for sedation during non-invasive procedures in radiology [36].

Due to non-availability of pediatric FOB, Kundra P et al. have used adult FOB, which was passed through one nostril and endotracheal tube (ETT) through the other nostril and intubated the trachea with video camera assistance. This technique was used in two cases. First, in a 14-month-old girl with

congenital bilateral TMJ ankylosis along with micrognathia and glossoptosis with 5-mm mouth opening and secondly in, a 3-year-old male with post traumatic TMJ ankylosis with nil mouth opening. Since awake fibreoptic is not feasible in children, induction with inhalational agent like sevoflurane and maintaining spontaneous ventilation is an alternative method. But cessation of ventilation with sevoflurane, once the mask is lifted away, allows limited time for securing the airway. Hence, changeover to halothane or isoflurane after induction with sevoflurane would prevent rapid emergence and allow more time for establishing the airway. An alternative could be the use of a smaller-sized ETT as a nasopharyngeal airway to help in maintaining airway patency, administer oxygen, and also acts as a conduit for delivering anesthetic gases for maintaining adequate depth during the procedure [37].

Similarly, Fiadjoe J et al. applied the above technique for maintaining and securing the airway in a 5-year-old child with bilateral TMJ, mild retrognathia, and 5-mm mouth opening. In a 2.5-year-old child with right TMJ ankylosis and mild retrognathia, the presence of enlarged adenoids precluded the use of blind nasal intubation. After induction of anesthesia with halothane, adult FOB was introduced orally and the glottis visualized thereafter, a 4.5-mm uncuffed ETT was passed orally into the trachea [24].

Fluoroscopic assisted airway intubation was attempted by Varughese I et al. in five TMJ ankylosis patients with 100 % success rate. This technique was devised to overcome the non-availability of FOB [38]. Although successful, this procedure remains cumbersome and time-consuming with the added problem of radiation exposure. Its applicability in pediatric age group remains doubtful.

Dhasmana S et al. reported using midazolam 0.05 mg/kg plus fentanyl 3 µg/kg in bolus for blind nasotracheal intubation under conscious sedation in adults when a FOB is not available [39]. This technique may not be appropriate for pediatric patients.

A novel technique of using a gum elastic bougie in a case series of three pediatric patients with TMJ ankylosis was described by Arora et al. After inducing the patients with thiopentone, the airway was maintained with air, oxygen, and halothane. Thereafter, an ETT was passed through one nostril and advanced till maximum intensity breath sounds could be heard. Another ETT was passed via the other nostril to deliver anesthetic agents and maintain depth of anesthesia. Finally, a gum elastic bougie was introduced via the first ETT and advanced into the glottis. This method proved to be easier and less traumatic compared to the conventional blind nasal intubation [40].

Preoperative Assessment

Detailed history and physical examination is a must. Children with OSA usually snore, and the absence of history of snoring

makes the diagnosis of OSA unlikely. Presence of symptoms such as daytime sleepiness, disturbed night sleep, mouth breathing, lack of concentration in school, and neurobehavioral symptoms are suggestive of OSA. Preoperative investigation like polysomnography in patients with history suggestive of OSA can help in ascertaining the degree of severity of OSA and also for differentiating primary snoring from OSA. It is also important to find out if the child was using any airway support devices like oxygen supplementation, CPAP etc. at home. These devices may be used in the postoperative period for supporting airway during recovery from anesthesia.

OSA in children has been more studied in adenotonsillar hypertrophy, where not all patients undergo sleep studies. The diagnosis of OSA is more often based on clinical findings. The presence of a compensatory metabolic alkalosis on blood gas analysis indicates a degree of chronic hypercarbia. Whenever preoperative polysomnography has been performed, in addition to Apnea Hypopnea index (AHI), the nadir and duration of oxyhaemoglobin desaturation, and also the peak end-tidal carbon dioxide measurement have to be considered for assessing the severity of OSA. Chronic hypercarbia and thereby increased pulmonary pressures resulting in cor pulmonale may develop in patients with nadir episodes of oxygen desaturation reaching 70 % during sleep. An electrocardiogram, echocardiogram, and further evaluation by a cardiologist if needed should be done before an elective surgery. As compared to adults cardiopulmonary abnormalities are less often seen in children [26]. Children with severe OSA due to TMJ ankylosis require distraction osteogenesis (DO) surgery to elongate the length of the mandible and prevent the tongue from obstructing the airway [13]. Due to difficulty in feeding, often these children are undernourished. Routine hemogram and serum electrolytes usually suffice in the absence of any other positive finding.

Premedication

Extreme precaution has to be exercised while prescribing sedative premedication to such patients. It should preferably be avoided as it may aggravate airway obstruction. Sedatives, opioids, and even anesthetic agents reduce the pharyngeal muscle tone and may lead to upper airway collapse. They also cause reduced arousal and ventilator response to hypoxemia and hypercapnia [16]. If premedication has to be given, then patient has to be continuously monitored. Sometimes, the residual effect of premedication may even be seen in the postoperative period in the form of respiratory depression [41, 42].

Use of antisialogogue medication dries up the oral secretions making visualization by FOB easier. Nasal decongestant drugs xylometazoline drops or adrenaline soaked gauze pieces helps in decreasing the nasal mucosal vascularity. This decreases the chances of bleeding during FOB guided nasotracheal intubation.

Perioperative Management

As children will not allow awake FOB, induction with inhalational anesthesia and spontaneous breathing is usually the method of choice. Mask ventilation may be difficult and chin lift and jaw thrust may not always improve airway patency.

In order to maintain adequate depth of anesthesia, a flexible soft nasopharyngeal airway can be introduced through the less patent nare. Oxygen and anesthetic gases can be administered through this airway and gentle mask ventilation can also be attempted in small children after occlusion of the other nostril. Thereafter, FOB-guided nasotracheal intubation can be attempted through the more patent nostril after ensuring adequate depth of anesthesia.

Combined intravenous and inhalational induction can also be used for securing the airway. Slow intravenous induction with propofol followed by maintenance with an inhalational agent can help to maintain spontaneous ventilation. Short-acting analgesics like remifentanyl can also be used for induction and for providing analgesia during the procedure. This should be accompanied by use of nonsteroidal anti-inflammatory drugs such as paracetamol and ketorolac half an hour prior to end of the procedure to provide postoperative analgesia.

Use of muscle relaxants also have been suggested by some for providing good intubating conditions; however, the anesthetist should have sufficient expertise in securing the airway prior to administering a non-depolarizing muscle relaxant in these children with a difficult airway [33].

Older children (>10–12 years of age) may agree to take topical local anesthetic spray, nebulization, or gargle [43]. An awake FOB-guided intubation may be attempted in these children. Use of a cuffed tube is preferable (microcuffed tubes for smaller children) as this would prevent the child from aspirating blood collected in the oral cavity during the procedure. In addition, whenever possible oropharyngeal packing should be done. North facing preformed tubes such as the RAE (Ring-Adair-Elwyn) help to secure airway without intruding on the surgical field.

Postoperative Management

Extubation of trachea should always be carried out when the patient is fully awake. Patients need post operative monitoring especially the ones with OSA. Usually, following the surgery, the patients develop adequate mouth opening. Therefore, they can be reintubated in case the need arises. If patient was on any airway support devices, it may be used in the postoperative period as well. Postoperative analgesia can be provided with round the clock non-steroidal anti-inflammatory drugs so that the patient has minimal postoperative pain.

Compliance with Ethical Standards

Conflict of Interest The authors declare that they have no conflict of interest.

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