Multidisciplinary Approach in Treating Undiagnosed Severe Temporo Mandibular Joint Ankylosis : A Case Report

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Background: Temporo Mandibular Joint (TMJ) ankylosis refers to bone or fibrous adhesion of the anatomic joint component and the ensuing loss of their function. The TMJ forms the very cornerstone of craniofacial integrity and hence its ankylosis in growing children adversely affects the growth and development of the jaws and occlusion. Difficulty in mouth opening and mastication, poor oral hygiene and rampant caries pose a severe physical and psychological burden in the tender minds of children.
Patient and Method: Bony ankylosis on the right TMJ in a female patient was not diagnosed until the patient reached her early teens, at which the condition was treated by bony fusion release on the right condyle. We managed the patient for further orthognathic surgery (Le Fort I Ostoeotomy and Bilateral Sagittal Split Osteotomy) to correct the skeletal deformity.
Result: In collaboration with the Orthodontist for pre-surgery and post-surgery orthodontic treatment, and Physiotherapist for mouth opening and masticatory muscles exercises, a good functional and aesthetic result was achieved.
Summary: Multidisciplinary approach in treating severe TMJ ankylosis is mandatory to achieve the optimum results. Awareness among all plastic surgeon and dentist involved in the treatment of craniofacial pathologies in children must be build to allow early diagnosis and treatment.
Keywords: *TMJ ankylosis, orthognathic surgery, pre and post-surgery orthodontic treatment*

Latar Belakang: Ankylosis pada TMJ didefinisikan sebagai adhesi tulang atau jaringan lunak pada sendi TMJ yang menyebabkan kerusakan fungsi persendian. TMJ memegang peranan penting pada integritas kraniofasial, sehingga ankylosis yang terjadi pada masa tumbuh kembang dapat mengganggu oklusi dan pertumbuhan rahang. Kesulitan pasien untuk membuka mulut dan mengunyah, disertai higiene oral yang buruk dan karies gigi, dapat menyebabkan gangguan fisik dan psikologis pada anak-anak.
Pasien dan Metode: Kami melaporkan kasus pasien anak perempuan dengan ankylosis tulang pada regio TMJ kanan yang tidak terdiagnosis hingga usia remaja. Operasi pertama yang dilakukan adalah melepaskan fusi tulang di kondilus TMJ kanan. Selanjutnya dilakukan operasi ortognatik yaitu Le Fort I Osteotomy dan Bilateral Sagittal Split Osteotomy untuk memperbaiki deformitas pada tulang-tulang wajah.
Hasil: Dengan tatalaksana multidisiplin bersama sejawat Orthodontist dan Physioterapist untuk perawatan gigi geligi pra dan pasca operasi, serta latihan otot-otot pengunyah, pasien menunjukkan hasil operasi yang baik dari segi fungsional dan estetik.
Ringkasan: Pendekatan multidisiplin untuk penanganan ankylosis TMJ yang berat mutlak diperlukan demi mencapai hasil terapi yang optimal. Kepedulian yang tinggi harus dibangun di kalangan dokter bedah plastik dan dokter gigi yang terlibat penanganan kasus kraniofacial sehingga penegakan diagnosis dini dan tatalaksana dapat dilakukan dengan segera.

Ankylosis is a Greek terminology meaning 'stiff joint'. It can be defined as inability to open mouth due to either a fibrous or bony union between the head of the condyle and the glenoid fossa. There is an intra-capsular union of the disc-condyle complex to the temporal articular surface that restricts mandibular movements, including the fibrous adhesion or bony fusion between condyle, disc, glenoid fossa and eminence. [1]

Temporo Mandibular Joint (TMJ) ankylosis is most commonly associated with trauma (13-100%), local or systemic infection (10-49%), or systemic disease (10%), such as ankylosing spondylitis, rheumatoid arthritis and psoriasis. Ankylosis can also occur as a

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result of TMJ surgery. The treatment of TMJ ankylosis poses a significant challenge because of technical difficulties and a high incidence of recurrence.[2]

Though TMJ ankylosis is one of the most common pathologies afflicting the facial skeleton, it is also the most over-looked and under-managed problem in children. Ankylosis in children is a serious and disabling condition. Functional sequelae include limited mandibular range of motion, impairment of speech, difficulty of mastication, poor oral hygiene, rampant caries, malocclusion and acute compromise of the airway. Furthermore, ankylosis in children may result in under-development of the mandible and aberrant facial growth.[3]

PATIENT AND METHODS

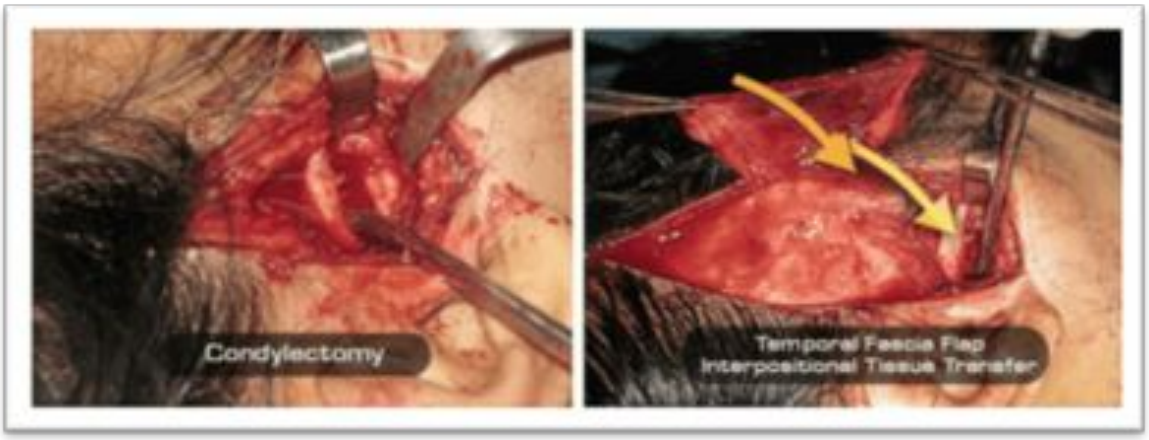
A 16 years old girl reported with the complaint of inability to open her mouth wide (Fig 1). History revealed that she might have an unrecognized episode of trauma to her chin when she was about 2 or 3 years old. She had been consulted a local doctor in Lampung who seemingly missed the diagnosis. She had difficulty in mastication, so she can only eat blenderized food and drink milk or fluid. Her oral hygiene was also poor with lots of rampant caries. When she was examined at our out-patient clinic, her mouth opening was as little as 5 mm, and her TMJs were tender on palpation. She was also showed a significant facial disturbances described as bird face deformity and mandible hypoplasia. Radiographic evaluation comprised of a facial CT-Scan which revealed mandible hypoplasia and maxillo-mandibular bony fusion of the right TMJ. Based on all these findings, a diagnosis of TMJ bony ankylosis and mandible hypoplasia was confirmed (Fig 2).

Fig 2. Mandible hypoplasia (left) and maxillo-mandibular bony fusion of the right TMJ (right).

The patient's first surgery procedure was done in Cipto Mangunkusumo Hospital on August 2010. She underwent condylectomy procedure at the right side of TMJ, arthroplasty and interpositional tissue transfer to the TMJ with temporalis superficial fascia flap (Fig 3).

For the next 8 months patient was managed by the Orthodontist to perform the pre surgery orthodontic treatment. The final orthodontic goal is to position the teeth ideally in the context of planned surgical procedure and facial aesthetics. Ideal position of the teeth in the arrangement within the basal bone, allows the surgeon to position the basal bone

Figures 1. Patient's first examination on June 2010. Her mouth opening was as little as 5 mm, and her TMJs were tender on palpation. She was also showed a significant facial disturbances described as bird face deformity and mandible hypoplasia.



Figures 3. Condylectomy procedure at the right side of TMJ, arthroplasty and interpositional tissue transfer to the TMJ with temporalis superficial fascia flap.

within the proper anatomical relationship to the cranial base and between maxilla and mandible. This will in turn result in ideal and stable skeletal, occlusal and neuromuscular relationship and in physiological TMJ relationship to the cranium.

Before the operation procedure, a clinical examination, cephalometric evaluation, and diagnostic models must be completed, and the surgery was planned with this new set of data. This new diagnostic information should be obtained within two weeks of the surgical date, as to prevent ongoing orthodontic changes from having a significant impact on the surgical plan (Fig 4).

The following orthognathic surgery treatment plan was developed : Le Fort 1 Osteotomy, Bilateral Sagittal Split Osteotomy (BSSO)

Figures 4. After the right condylectomy procedure, patients mouth opening was about 2.5 cm. Then she was managed for about 8 months by the Orthodontist to perform the pre surgery orthodontic treatment.

Figure 5. Cephalometric analysis.

and Genioplasty, to correct the jaws and facial deformity.

Orthognathic Surgery Treatment Plan: (a) Maxilla: Le Fort 1 Osteotomy, the maxilla will be moved backward 4 mm at ANS, and will be rotated anterior up 4 mm and posterior up 2 mm at PNS (clockwise rotation). The pivoting point is around the first molar. Superior impaction of maxilla on the left side by 3 mm will be done to correct the maxillary occlusal; (b) Mandible: Bilateral Sagittal Split Osteotomy (BSSO), the mandible will be moved forward 9 mm at right side and 2 mm backward at the left side, therefore the chin will be moved forward by 9 mm at pogonion and by 7 mm at the right side. Tooth extraction of 18,28,38,48 and 45 impacted; (c) Augmentation Genioplasty, augmentation genioplasty will be done by 7 mm forward at pogonion. Augmentation will be done using costal cartilage graft.

The following pictures show the intraoperative procedures (Fig 6):

Figures 6. Intra Operative Pictures: Le Fort 1 Osteotomy, Bilateral Sagittal Split Osteotomy (BSSO) and Augmentation Genioplasty

After the operation, maxillo-mandibular fixation was maintained for 5 days and the patient was discharged from hospital 5 days after surgery. Operation results after the orthognathic procedures, showed significant improvements in mouth opening, masticatory function and facial deformity (Fig 7).

Then she was referred back to the Orthodontist for post-surgical orthodontic treatment to maintain the optimal and most stable occlusal relationship. Retainers then will be used to ensure the stability of the teeth until the periodontal supporting mechanism has reorganized in the new position. Physiotherapy treatment plan will be developed for this patient to improve the functions of masticatory muscles and prevent relapse of the jaws position.

Figure 7. Pre and post operative from AP view (upper left), Radiology before and after operation (upper right), pre and post operative from lateral view (lower left, lower right)

DISCUSSION

The phrase "temporomandibular joint disorders" is a broad umbrella diagnosis that encompasses functional disturbances and anatomical disorders of the TMJ. Functional disturbances are characterized by restrictive active motions in the context of normal passive range of motion. TMJ ankylosis, bony or fibrous union between the articular surfaces of the joints, is relatively rare, but can be extremely morbid and disabling. Although it can occur in any age group, children are at higher risk because of anatomical and physiologic differences at the joints.[3]

Functional sequelae include limited mandibular range of motion, inability to form an oral seal, interference with mastication, and difficulty with nutrition and oral hygiene. Furthermore, ankylosis in children may result in underdevelopment of the mandible and aberrant facial growth.[4]

Causes of TMJ disorders are varied, including: (a) Organic disease that may directly affect the joint, i.e: rheumatoid arthritis, osteoarthritis, degenerative joint disease, psoriatic arthritis and ankylosing spondylitis; (b) Local or syste-mic infection may results in capsulitis, osteomyelitis and suppurative arthritis; (c) Trauma, can result in fracture, ligamentous injury, and anterior joint dislocation; (d) Injury from surgery, radiation therapy or embolization therapy may result in soft tissue scarring or fibrosis that can restrict

the motion of the joint; (e) Bony and soft-tissue cancers may directly invade the joint or externally restrict mandibular excursion. The most common tumors chondromas, osteomas, osteochondromas, chondroblastomas, and fibrous dysplasia; (f) Congenital and developmental i.e: condylar agenesis or hypoplasia, condylar hyperplasia, craniofacial microsomia, and coronoid hypertrophy.

The most morbid and disabling manifestation of temporomandibular joint dysfunction is immobility caused by ankylotic fusion of the mandible to the cranial base or zygoma. Studies have shown that restricted mouth opening in TMJ ankylosis creates not only functional and aesthetic problems, but also its sequelae including poor oral hygiene and rampant caries, facial asymmetry, mandibular micrognathia and bird face, malocclusion and absence of condylar movements. The clinical manifestations depend to a large extent on the age at the time of onset, duration, anatomical location and involvement of one or both joints (unilateral or bilateral).[7]

The hallmark of temporomandibular joint disorders is decreased inter-incisal opening (typically <15 mm), with or without pain. In its severest form, it can be extremely incapacitating, resulting in limited mouth opening, malocclusion, interference with mastication, inability to form an oral seal, and thus difficulty achieving adequate nutrition and maintaining oral hygiene.

Temporomandibular joint ankylosis in the pediatric patient results in restricted mandibular growth and retrognathism. Specific manifestations are contingent on laterality: in unilateral ankylosis, the chin is deviated ipsilaterally/posteriorly because of mandibular ramal shortening on that side; in bilateral cases, there is mandibular retrusion without asymmetry. Unilateral ankylosis typically results in a crossbite, whereas the more severe mandibular underdevelopment in bilateral cases typically results in micrognathia and an anterior open bite.[8]

In some cases, particularly in children, the mandibular condylar neck is shortened and the coronoid process extends above the level of the zygomatic arch. Pressure from lips and tongue-thrusting (when trying to form an oral seal) may adversely mold dentition. However, overall functional occlusion is rarely affected to any significant degree because of the adaptive capacity of the alveolus, which remodels to allow for dental repositioning.

Untreated unilateral ankyloses may result in bilateral disease, as restricted movement eventually causes a fibro-osseous union on the contralateral side. The severity of symptoms and specific sequelaeare highly dependent on age at the time of onsetof ankylosis, because injury in the growing child may have a pronounced effect on growth and development.

Temporomandibular joint disorders can be broadly categorized into functional disturbances (e.g., tetany, myofascial pain syndrome) and anatomical abnormality. Anatomical disorders are subdivided into soft-tissue or skeletal defects, depending on which part of the anatomy (e.g., musculature,mucosa, and glands, versus joint and skeleton) is principally affected.

Another TMJ ankylosis classification was according to Sawhney's, he classified TMJ ankylosis into four different types: (1) Type I, when there was minimal bony fusion, but extensive fibrous adhesions around the joint; (2) Type II, when there was more bony fusion, especially at the outer edge of the joint surface, but no fusion within the more medial area of the joint; (3) Type III, when there was a bridge of bone between the mandible and the temporal bone; and (4) Type IV, when the joint was replaced by mass of bone.

Preoperative assessment in TMJ ankylosis included a thorough history and physical examination to determine the ankylosis characteristics according to age of onset of the ankylosis, etiology, involvement, ankylosis type, treatment, maximal incisal opening (MIO) and presence of facial asymmetry.

Surgical procedure of the TMJ was done by the pre-auricular approach described by Ellis and Zide under general anesthesia. After exposure and identification of the site of the ankylosis, aggressive excision of the fibrous and/or bony mass was carried out with round burs and chisels until the mandibular movements were achieved. Next the glenoid fossa was recountered as necessary. For all three surgical procedures, coronoidectomy on the ipsilateral side was performed only if the passive maximal

opening was less than 35mm. For the gap arthroplasty, in addition to this procedure, a gap of at least 15mm was created between the glenoid fossa and the mandible using interpositional tissue transfer to the TMJ with temporalis superficial fascia flap. The temporalis superficial fascia flap is an autogenous graft that has advantages of close proximity to the TMJ minimal surgical morbidity and successful clinical results. It was found to be a valuable option for TMJ ankylosis reconstruction.

Le Fort I osteotomy, with or without segmentation, can be used to correct a variety of maxillofacial deformities in all planes. When the maxilla is to be moved anteriorly or inferiorly, consideration should be given to placing bone grafts in the gaps that invariably will be present once these movements are accomplished. Once all interferences are removed, the anticipated movements are measured against external landmarks. The splint is then placed and maxillo-mandibular fixation (MMF) is accomplished with the splint in place by utilizing wire or elastics to the surgical hooks on the orthodontic wires. It is important at this stage to utilize the mandible as a point of reference. With the maxilla in the proper position, the next step is to accomplish fixation. In the maxilla, this is usually done with 2 mm plates. Usually two L-shaped plates are utilized per side and are positioned at the zygomaticomaxillary buttress and the piriform rim buttress areas.

After the fixation is completed, the new maxillary position is checked with the external landmarks. It is important to check both sides of the midline to ensure that there is no canting of the maxillary plane. Once the surgeon is satisfied with the new maxillary position, the incisionis closed, usually with a 3–0 or 4–0 resorbable suture.

The procedure of choice for treatment of mandibular prognathism is a sagittal splitosteotomy. As previously discussed, surgery should be undertaken only after dental compensations are eliminated with presurgical orthodontics and, preferably, after mandibular growth is complete. In addition, advancement genioplasty and submental or submandibular liposuction or lipectomy should be considered to improve the patient's overall appearance. Treatment of isolated mandibular deficiency usually involves mandibular advancement. Bilateral sagittal split osteotomy (BSSO) with rigid fixation is the most frequently performed procedure. The BSSO is a very versatile operation in that it can be used both for mandibulara advancement and for mandibular push-back. It lends itself to rigid fixation, which, in turn, means that the patient will not require maxillomandibular fixation. The BSSO is an excellent operation for mandibular advancement to about 12 mm. If the advancement required is greater, other options should be considered, because there will be minimal to no overlap between the proximal and distal segment, creating a much greater potential for relapse.

Chin reconstruction occupies an important aspect of facial balance and an object of admiration for centuries. Facial contouring surgery is an important part of a plastic surgery practice. Its goal is the correction of the bony tissues of the face to obtain facial harmony. This harmony is mainly determined by the size, shape, position, and proportion of the chin in respect to the other facial elements. The indications for genioplasty include positioning of the chin in the facial midline and alterations in the vertical or horizontal position of the chin in order to achieve better facial balance. A geniopalasty can be done as a single procedure or in conjunction with orthognathic surgery as part of an overall treatment plan. In this patient, we applied the genioplasty augmentation using costal cartilage graft.

SUMMARY

A certain number of patients with facial cosmetic complaints have skeletal and occlusal abnormalities that are the source of, or greatly contribute to, their chief complaint. Thus, it is important for the plastic surgeon evaluating these patients to be familiar with the tools used to evaluate the orthognathic surgery patient. The correct diagnosis of the etiology of the patient's problem is of paramount importance for adequate treatment and to achieve optimal results.

The aim of this article is to present an overview based on a case report, of efficient management strategies in multidisciplinary approach, including the pre-surgery orthodontic treatment, in treating severe TMJ ankylosis, so as to increase its awareness among all plastic surgeon and dentist involved in the treatment of craniofacial pathologies in children.

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