

TREACHER COLLIN'S SYNDROME WITH TEMPOROMANDIBULAR JOINT
ANKYLOSIS-A VARIANT- SURGICAL CORRECTION BY GAP ARTHROPLASTY
- A CASE REPORT WITH THE REVIEW OF LITERATURE

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ABSTRACT--Temporomandibular joint ankylosis is a bony or an intracapsular fibrous fusion of the mandibular condyle and the glenoid fossa complex. The patient experiences severe cosmetic deformity along with restricted mouth opening, difficulty in swallowing. The facial deformity due to severe ankylosis makes a psychosocial problem to the patient. The prompt treatment to relieve the ankylosis is a challenging task to the surgeon. We report a case of 24 year old female came to the ENT out patient department with the complaints of inability to open the mouth, noisy breathing-chronic stridor and facial deformity since birth. On examination patient is having trismus, hypoplastic mandible, protruded upper jaw, malocclusion of tooth in both jaws and diagnosed as traecher Collin syndrome (variant) with ankylosis of temporomandibular joint. We performed GAP arthroplasty with external fixator. On follow up the patient after 6 months, patient was relieved of noisy breathing, trismus.

KEYWORDS -- Gap arthroplasty, Temporomandibular joint ankylosis, Trismus, Stridor.

1,INTRODUCTION

Temporomandibular joint ankylosis (TMJ) is a rare disorder. It occurs due to the fusion of mandibular condyle at the base of the cranium. The etiology is mainly trauma, systemic diseases and can be congenital too. TM joint is a synovial joint which is essential for mastication, speech



and is formed between condyle of mandible below and the articular fossa of the temporal bone above¹. Temporomandibular joint ankylosis is cosmetically a disfiguring condition and it is an ankylosis between bony fusion of mandibular condyle and glenoid fossa ². We report a case of temporomandibular joint ankylosis which was treated by Gap Arthroplasty.

2,CASE REPORT

A 24 year old female presented with inability to open the mouth, dysphagia, noisy breathing, inability to articulate the speech during development, facial disfigurement since birth (Figure 1). Inability to open the mouth (trismus) is congenital in origin. Dysphagia more for solids not associated with regurgitation of food and fluids, patient had noisy breathing (stridor) with indrawing of chest while sleeping for the past 12 years.





Figure 1 Figure 2

On examination patient was diagnosed as a rare variant of traecher Collin syndrome based on hypoplastic mandidle, protruded upper jaw, malocclusion of tooth in both jaws, congenital trismus with ankylosis of temporomandible joint left side (Figure 2).







Figure 3 Figure 4

Patient radiologically investigated with orthopantogram (Figure 3), Xray skull lateral oblique view (Figure 4), xray Paranasal sinuses-Water's view (Figure 5) suggestive of micrognathia, severe dental occlusion, partial craniostenosis (sagittal sutures not seen), left temporomandibular joint ankylosis. Xray neck lateral view was normal (Figure 6). Case is planned for left GAP arthroplasty for the relief of trismus and stridor.





Figure 5 Figure 6



Under the flexible fibrooptic bronchoscopic guidance, Rail road technique of intubation done with flexometallic endotracheal tube. A vertical preauricular incision made from 2 inch above the superior border pinna to the ear lobule (Figure 7). Under the blunt, horizontal dissection, temporalis fascia with muscle separated with preservation of facial nerve. Left temporomandibular joint exposed (Figure 8).



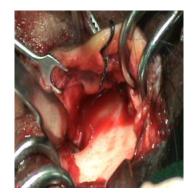


Figure 7 Figure 8

Joint capsule is intact. No meniscus. Head of the mandible is flattened and protruded condyle removed by mallet and gouge along with microdrill (Figure 9, 10). GAP arthroplasty done. Gapping of 2 cms is achieved (Figure 11). Poly Dioxanon Sheath (PDS) kept between glenoid cavity and coronoid process (Figure 12). External pin fixator done between zygomatic arch to angle of mandible for post operative distraction (Figure 13). Wound closed in layers with preservation of facial nerve (Figure 14).







Figure 9 Figure 10

Dressing done. Patient kept in Intensive Care Unit, along with flexometallic endotracheal tube for first 48 hours and then removed. On post operatively 3rd day, patient able to take food and fluid, speech improved, stridor relieved, able to protrude the tongue outside. Sutures removed on the 7th day.







Figure 11 Figure 12 Figure 13



By daily distraction done with external pins and a wide mouth opening is achieved. At the end of 6 months external pins removed and mouth opens 6cms widely between jaws (Figure 15,16).







Figure 14 Figure 15 Figure 16

3, DISCUSSION

Temporomandibular joint ankylosis is a very distressing condition where TM joint is replaced by scar tissue. It is classified based on ³

- 1. Combination of location as intra articular or ectra articular
- 2. Type of tissue involved as bone, fibrous or fibro- osseous
- 3. Extent of fusion as complete or incomplete

Intra-articular ankylosis is due to trauma or infection, but extra-articular ankylosis occurs by a variety of disorders including myogenic, neurogenic and inflammatory processes, bone and soft tissue tumors ⁴ These patients will have severe difficulties for swallowing, mastication and speech

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and present with trismus. The ankylosis is most commonly occurs due to maldevelopment of the

meniscus and its existence.

Surgical corrections of TMJ ankylosis are Gap arthroplasty, interpositional arthroplasty,

excision and joint reconstruction with grafts, and total TMJ replacement with joint prosthesis. The

main disadvantage of all the procedures is chance of recurrence ⁵. The principle of gap arthroplasty

is to minimize the rate of ankylosis process by increasing the distance between ascendant ramus

to the cranial base. If the distance is small, it can lead to recurrence.

The induction of anaesthesia to Temporomandibular joint ankylosis patients is a challenging

task for securing the airway. The adequate monitoring facilities are mandatory. The best safest

technique in securing the airway is by using a nasal fiberoptic assisted intubation with the patient

awake and under local anesthesia ⁶. When Temporomandibular joint ankylosis occurs in a child, it

causes the drastic effects on the future growth and development of facial bones, teeth, jaws and its

union. This in turn leads to extreme negative influence on the psychosocial disturbances, as it

causes facial deformity and it worsens with future growth ⁷.

4, CONCLUSION.

Comparing to the other surgical procedures for Temporomandibular joint ankylosis, GAP

arthroplasty is less invasive, less surgical time comfortable post operative period. The outcome of

the surgery also depends on continous mouth opening exercises for atleast 6 months with external

pin¹. In this patient, chronic stridor of 12 years was totally relieved and the daily day today

activities improved well with adequate opening of mouth. Interposition of synthetic material Poly

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Dioxanon Sheath (PDS) between ossified raw area is preventing the recurrent ankylosis during the

follow up period.

In this case, as the GAP arthroplasty is a rare surgery done for few decades with review of

literature shows exclusion of neurogenic tissues, soft tissues, inflammatory tissues is important. In

comparison of 7 references (cited in this article) with a review of literature GAP arthroplasty with

the insertion of synthetic material Poly Dioxanon Sheath (PDS) is rare, which is highlighted in this

case. In this patient Temporomandibular joint ankylosis is congenital in origin, unilateral left side

where GAP arthroplasty done with Poly Dioxanon Sheath (PDS) with good improvement which

can be used for other acquired Temporomandibular joint ankylosis also.

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