RESEARCH ARTICLE

# Correction of hypoplastic mandible in non-syndromic temporomandibular joint ankylosis patients with distraction osteogenesis in Mayo Hospital Lahore: A descriptive study

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## Abstract

**Objective:** To evaluate the efficacy of simultaneous gap arthroplasty and distraction osteogenesis in the treatment of non-syndromic temporomandibular joint ankylosis patients with hypoplastic mandible.

**Methods:** The descriptive prospective study was conducted at the Department of Oral and Maxillofacial Surgery, King Edward Medical University / Mayo Hospital, Lahore, Pakistan, from January to December, 2016, and comprised patients having temporomandibular joint ankylosis with hypoplastic mandible. Interpositional gap arthoplasty, osteotomy and application of distractor were done in all the patients. Preoperative and postoperative data was collected and analysed using SPSS 12.

**Results:** Of the 63 patients, 29(46%) were males with a mean age of 18.48±2.32 years and 34(54%) were females with a mean age of 16.41±3.21 years. Overall mean age was 17.37±3 years (range: 9-24 years). Of the total, 20(32%) patients were given a single distractor on ramus per side, while 43(68%) were given double distractors (ramus + body) per side. Lengthening was achieved in all (100%) patients both in ramus and body. Only 1(1.6%) patient presented with a complication during the distraction period, and the case was successfully managed surgically.

**Conclusion:** Distraction osteogenesis is a promising treatment option for the correction of mandibular deformities as a result of temporomandibular joint ankylosis.

**Keywords:** Distraction osteogenesis, TMJ ankylosis, Interpostional gap arthroplasty, Mandible, Osteotomy. (JPMA 70: 24; 2020). **https://doi.org/10.5455/JPMA.297738**

## Introduction

Distraction osteogenesis (DO), or callus distraction, is the generation of viable bone by the gradual separation of osteotomised bone segments. The force and tension applied to the separated bone-ends signal the body to form new bone in the gap between the bony edges. This force and tension applied also create tension in the surrounding soft tissues which initiates a sequence of adaptive changes termed distraction histogenesis (DH). With steady distraction taking place in the bone, it also initiates histogenesis in neighbouring tissues, including skin, neurovascular tissue, muscles, ligaments and periosteum. This phenomenon of histogenesis permits large skeletal movements with very little chance of relapse compared to acute orthopaedic corrections.[1]

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DO was first introduced in 1905. In 1927 a similar study was published, but the idea of distraction was popularised in 1951. In the craniofacial region, the first successful distraction of the human mandible was done in 1989.[2]

Ankylosis is a Greek term meaning 'stiff joint'.[3] Temporomandibular joint (TMJ) ankylosis is the most common cause of facial asymmetry due to hypoplastic mandible in non-syndromic individuals. Longstanding TMJ ankylosis leads to gross facial asymmetry as well as respiratory and psychological abnormalities.[3,4] Common causes of TMJ ankylosis are trauma and infection.[3,5] TMJ ankylosis is common in our part of the world due to lack of awareness, facilities to treat condylar fractures, and infections at primary or secondary healthcare levels.[6] Even at tertiary healthcare centres, TMJ ankylosis treatment cannot be done due to lack of maxillofacial surgeons.

Historically, TMJ ankylosis patients are treated with gap

arthroplasty (GA)[3,7] and interposition of fascia or muscle, and reconstruction is done with costochondrol bone grafts.[7-9] Costochondral graft is used for reconstruction because of its growth capacity and biological similarities to condyle of mandible.[9]

Due to the unpredictable nature of the graft, unsatisfactory results, such as resorption of the graft, facial asymmetry, growth disturbance and re-ankylosis, are often encountered. Even if the costochondral graft is successful, donor-site morbidity is inevitable.[9]

DO is an alternative treatment modality for the correction of mandibular deficiency due to TMJ ankylosis. Previously, studies have been conducted for the correction of vertical height deficiency of the ramus and horizontal body lengthening of the mandible, causing facial deformities due to TMJ ankylosis in non-syndromic patients.

The pioneers of bi-directional osteodistraction in the mandible, to lengthen both ramus and body of the mandible were Molina and Ortiz-Monasterio.[10]

DO following interpositional gap arthroplasty (IGA) is a far superior method for the correction of TMJ ankylosis than conventional bone grafting due to its reliability and predictability.[11] Moreover, with the use of this technique, bone grafting and second-stage jaw correction surgery can be avoided, which decreases surgical morbidity.

At present no study is available at the national level in Pakistan about the use of bidirectional DO for the correction of facial skeleton abnormalities due to TMJ ankylosis. The current study was planned to evaluate the efficacy of simultaneous GA and DO in the treatment of non-syndromic TMJ ankylosis patients with hypoplastic mandible.

## Patients and Methods

The descriptive prospective study was conducted at the Department of Oral and Maxillofacial Surgery, King Edward Medical University / Mayo Hospital, Lahore, Pakistan, from January to December, 2016, and comprised patients having TMJ ankylosis with hypoplastic mandible.

After approval from the institutional ethics review board, the sample size was calculated using 90% confidence level, 10% margin of error and by taking expected mean change in ramus length after 6 months post-operatively as 6.75±1.51 (35.98%)2 respectively. The sample size was calculated using the given formula adopted by World Health Organisation (WHO) studies.[12]

$$n = \frac{z^2_{1-\alpha/2} P(1-P)}{d^2}$$

Z: Confidence level, which is 90% = 1.645
P: Prevalence = 6.75±1.51%
d: Margin of error = 10%

Using non-probability purposive sampling technique, patients aged 5-25 years having hypoplastic mandible due to TMJ ankylosis were included. Those having TMJ ankylosis due to underlying pathology, medically compromised patients and patients who had metal allergy were excluded. After informed consent was taken from the patients or their parents / guardians, detailed history, clinical examination, photograph and radiograph (orthopantomogram, lateral cephalogram [L-Ceph] and posteroanterior cephalogram [PA-Ceph]) of all patients were obtained. Cephalometric analysis of every patient was done to find out mandibular asymmetry due to vertical deficiency or horizontal deficiency or both; required vertical bone lengthening or horizontal bone lengthening or both; and planning of future IGA.

IGA, osteotomy and the application of distractor was done through extra-oral approach by the consultant. Pre- and post-operative data was collected. Following IGA and osteotomies on both sides, the distraction appliances were adapted. Some patients were given a single distractor on ramus per side while others were given double distractors (ramus + body) per side. During the latency period, L-Ceph and PA-Ceph were repeated to keep record of the mandibular length after the placement of the distracter and before the activation of the appliance. Activation of the appliance was done initially under supervision and then patients were given instructions on how to activate the appliance. In case of a child, the parents were instructed. When the desired lengthening was achieved, L-Ceph and PA-Ceph were repeated to measure the increase in the length of vertical ramus and horizontal body accordingly.

Data was analysed using SPSS 12. Quantitative variables, like initial length $T_1$, $L_1$ and final length $T_2$, $L_2$ after treatment, were documented as mean and standard deviation (SD). Paired sample test was used to determine the significant difference in pre- and post-treatment values. $P \leq 0.05$ was considered significant.

## Results

Of the 63 patients, 29(46%) were males with a mean age of 18.48±2.32 years and 34(54%) were females with a mean age of 16.41±3.21 years. Overall mean age was 17.37±3 years (range: 9-24 years) (Table 1). Of the total, 20(32%) patients were given a single distractor per side, while 43(68%) were given double distractors per side. Mean ramus length before distraction and after the excision of the ankylotic mass of all 63(100%) patients was 44.44±6.59mm on the left side and 45.11±7.06mm on the right side (range: 28-58mm).

Mean ramus length with DO after excision of the ankylotic mass of all patients was 54.51±6.16mm on the left side and 54.79±6.23 on the right side.

Mean increase in vertical ramus length was 10.04±2.35mm on the left side and 9.62±2.41mm on the right side (range: 5-15mm on both sides).

Of the total, 1(1.6%) patient had a complication regarding the desired distraction length. Bone lengthening was not achieved during the distraction phase due to failure of the distractor device for which surgical replacement of the distractors was done and the required lengthening was achieved.

**Table-1:** Descriptive statistics for age in relation to gender.

| \|'Age'\| | Gender | | Total |
|---|---|---|---|
| | Male | Female | |
| n | 29 | 34 | 63 |
| Mean | 18.48±2.32 | 16.41±3.21 | 17.37±3.00 |
| Minimum | 14.00 | 9.00 | 9.00 |
| Maximum | 24.00 | 23.00 | 24.00 |

**Table-2:** Descriptive mean increase in postero-anterior cephalogram (PA Ceph) and lateral cephalogram values of the patients for interpositional gap arthroplasty in Non-Syndromic temporomandibular joint (TMJ) Ankylosis patients.

| Comparison of the Rampus Lengthening and Body Lengthening Pre and Post Operatively | | | | |
|---|---|---|---|---|
| PA Ceph (Ramus lengthening) | | $T_1$ | $T_2$ | *p*-value |
| | Left | 44.44±6.59 | 54.51±6.16 | 0.01* |
| | Right | 45.11±7.06 | 54.79±6.23 | 0.01* |
| Lateral Ceph (Body lengthening) | | $L_1$ | $L_1$ | *p*-value |
| | Left | 83.76±7.01 | 98.63±6.83 | 0.01* |
| | Right | 84.60±6.53 | 98.81±6.77 | 0.01* |

*p*-value is <0.05, Significant result.

**Table-3:** Mouth opening (average).

| Pre-Op Mouth opening | Immediate Post-Op Mouth opening | Mouth opening 6 months after distraction | *p*-value |
|---|---|---|---|
| 3.3 ±1.2 mm | 33.07 ±1.81 mm | 36.8 ±2.7 mm | 0.01 |

Mean body length before distraction and after the excision of the ankylotic mass of 43(68%) patients was 83.76±7.01mm on the left side and 84.60±6.53 on the right side (range: 67-99mm).

Overall mean body length with DO after excision of the ankylotic mass was 98.63±6.83mm on the left side and 98.81±6.77 on the right side. Mean increase in horizontal body length was 14.88±2.80mm on the left side and 14.21±2.78mm on the right side (range: 9-20mm on the both sides (Table 2).

Mean mouth-opening pre-op was 3.3±1.2mm, while mean mouth-opening achieved immediate post-operatively was 33.07±1.81mm. At 6-month follow-up after DO, mean mouth-opening was 36.8±2.7mm (Table 3). Mean mouth-opening after distraction completion was 3.7±0.8mm.

## Discussion

Surgical intervention is the treatment of choice for the management of TMJ ankylosis and correction of mandibular deformity due to TMJ ankylosis. Surgical modalities include GA, IGA with costochondral graft[13] and IGA with DO.

GA is a surgical procedure in which liberal resection of the ankylotic mass is done and a gap of 1-1.5cm is created. In IGA, interpositioning of some autogenous or alloplastic material is done between condyle and temporal bone. The purpose of the interposed material is to lessen the possibility of recurrence and to establish a false joint.

Verneuil A was the first to use temporalis flap as an interpositional material in TMJ ankylosis. Warraich RA[14] established that interpositioning of temporalis fascia reduced post-operative pain and the need for prolonged physiotherapy, usually encountered with simple GA, and prevention of re-ankylosis.

In the light of all these methods, satisfactory mouth opening and prevention of re-ankylosis can be achieved, but for the correction of mandibular deformity due to TMJ ankylosis, a separate treatment option is needed.

Gillies introduced the use of autogenous costochondral graft for TMJ reconstruction. This graft is favoured because of its biological similarities to condyle of mandible and its capacity to grow.[9] But due to unpredictable nature of the graft, unsatisfactory results, such as resorption of the graft, facial asymmetry, growth

disturbance and re-ankylosis, are often encountered. Even if costochondral graft is successful, donor-site morbity is inevitable.[9]

DO is a reliable method for the correction of mandibular deformity as a result of TMJ ankylosis. In 1992, McCarthy et al[15,16] used DO for mandibular lengthening on four patients who had mandibular deficiency due to hemifacial microsomia and Nager syndrome. The study was able to achieve mandibular lengthening in all patients with the use of extra-oral distractors. The study is similar to the current study as far as achievement of lengthening is concerned. Our study differs from the previously mentioned study[15,16] as the number of patients involved in the earlier study was only four. Also, the current study had patients having mandibular deformity due to non-syndromic or non-congenital TMJ ankylosis, while the previous study involved patients of hemifacial microsomia and Nager's syndrome.[15,16]

A study[17] about the management of adult TMJ ankylosis patients with the use of transport DO arthroplasty (TDOAP) had 32 adult patients aged 18-61 years. It was able to correct facial asymmetry in all patients and was able obtain a mean increase in mandibular ramus length of 15.4mm, which is higher than mean increase in vertical ramus length of 9.62±2.41mm in the current study. It was able to achieve significant lengthening because the study was conducted on adult patients, and so greater lengthening was required for the correction of mandibular deformity due to TMJ ankylosis.[17] The current study had patients aged 9-24 years, and greater lengthening was not required for the correction of mandibular deformity.

A study[18] to evaluate DO for bilateral ankylosis of TMJ after osteoarthrotomy used DO after one year of IGA. It evaluated the technique on 20 patients aged 23-45 years. All patients were successfully treated and symmetrical chin position was reported in 100% cases. The study[18] is somewhat similar to our study as in both studies lengthening was achieved in vertical direction at mandibular ramus area. The earlier study is different as it only involved ramus lengthening whereas the current study involved body lengthening as well. The earlier study involved 20 patients, while our study had 63, and, of them, 43 cases underwent both body and ramus lengthening. Also, the study[18] used DO after one year of IGA, while in the current study, IGA and DO were applied simultaneously. Moreover, in our study, ages of the patients ranged 9-24 years, while the other study had it ranging 23-45 years.

A study[2] discussed the stability of the vertical ramus lengthening with DO in TMJ ankylosis patients. It was done on 8 non-syndromic TMJ ankylosis adult patients aged 17-33. These patients were treated successfully with GA and DO, lengthening was achieved up to 18mm. The study is comparable to the current study as both involved non-syndromic TMJ ankylosis patients. Vertical lengthening is also comparable as lengthening was achieved in ramus of the mandible up to 18mm in the earlier study[2] and up to 15mm in the current study. The study differs from the current study as it involved 10 adult patients aged 17-33 years, while the current study involved 63 patients aged 9-24 years.

A study[19] discussed the role of simultaneous GA and DO in the management of TMJ ankylosis with mandibular deformity in children. It was done on 6 non-syndromic TMJ ankylosis patients aged 7-10 years. Pre-operative mouth-opening was 0-6mm. Average post-operative mouth opening achieved after GA and DO was 28±4.19mm.[19] This study is comparable to the current study as both studies involved non-syndromic TMJ ankylosis patients in whom simultaneous GA and DO was done. Post-operative mouth opening is also comparable as in the earlier study post-operative mouth opening achieved was up to 36mm compared to 39mm in the current study. Our study differs from the earlier one[19] in terms of age range.

In terms of limitations, the patients were selected on the basis of their gross skeletal defect and were not part of any specific population. As is common with this disorder, there is no generalised scale or classification which encompasses all the varied pathologies, functional and / or cosmetic associated with it. Every individual presents with a different set of problems, be they cosmetic or functional. This pathology does not specifically appear in any specific set of population i.e. based on gender, age, race etc. Due to the long course of treatment and follow-up it has been seen that patients are not compliant after the initial surgery period when the gross skeletal defect is treated. Thus, follow-up and record maintenance of post-operative orthodontics is scarce. As is with classification or standardisation of the pathology, there is no standardised method or technique to rule out measurement errors. Measurements are done using basic equipment like vernier calipers or measuring scale.

In our study, all patients and measurements were assessed by three researchers to rule out individual bias. Only 6-month post-operative follow-up was available at the time of completion of the study period. We were not able to achieve ideal post-distraction occlusion due to limitation of the study period and, secondly, there was no orthodontics department available at the Mayo Hospital. We were not able to assess theatre charges for the procedures as the surgeries were done at the state-run Mayo Hospital, Lahore.

## Conclusion

Distraction osteogenesis for mandibular ramus and body lengthening with simultaneous temporalis fascia interpositional gap arthroplasty was found to be a useful and effective technique for the prevention of re-ankylosis and correction of mandibular deformity due to TMJ ankylosis. It shared all the advantages of autogenous bone grafting without the disadvantages of donor-site morbidity. At present it is considered the most pragmatic way to address both TMJ ankylosis and mandibular deformity at the same time.

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