

Efficacy of Distraction Osteogenesis in Management of Obstructive Sleep Apnea Secondary to Temporomandibular Joint Ankylosis

Shiv Shankar Agarwal¹, Nanda Kishore Sahoo², Sanjeev Datana³, Sujit K Bhandari⁴

ABSTRACT

Aim and objective: The aim and objective of this study was to evaluate the efficacy of mandibular corpus distraction osteogenesis (MCDO) in the management of obstructive sleep apnea (OSA) secondary to temporomandibular joint (TMJ) ankylosis.

Materials and methods: Treatment records of 15 patients (mean age 23.3 ± 5.4 years, 7 males and 8 females) comprehensively managed for severe OSA secondary to TMJ ankylosis with combined orthodontics and MCDO were studied objectively by comparing apnea-hypopnea index (AHI) scores and airway parameters, and subjectively by Epworth sleepiness scale (ESS) survey at pretreatment (T0) and 1-year postsurgery (T1). Change in skeletal parameters at T1 was evaluated by comparison of sella-nasion-point A (SNA), sella-nasion-point B (SNB), and point A-nasion-point B (ANB) angles. Change in AHI per millimeter advancement was also calculated.

Results: The mean AHI score at T0 and T1 was 41.71 ± 12.69 and 8.11 ± 3.25 , respectively (80.39% reduction, p -value < 0.001). The AHI reduction per mm distraction was 2.59 ± 0.52 . The success rate (postsurgical AHI less than 50% of pretreatment) was 100%, and the cure rate (AHI score < 05) was 80%. The mean ESS score at T0 and T1 was 18.60 ± 2.5 and 4.67 ± 1.49 , respectively (75.05% reduction, p -value < 0.001). The mean ANB at T0 and T1 was $11.6^\circ \pm 3.68^\circ$ and $3.4^\circ \pm 2.29^\circ$, respectively (72.89% reduction, p -value < 0.001). The mean posterior airway space (PAS) at T0 and T1 was 4.00 ± 1.46 mm and 10.00 ± 1.25 mm, respectively (182.44% increase, p -value < 0.001).

Conclusions: MCDO is an effective modality for the treatment of severe OSA secondary to TMJ ankylosis. Further studies are recommended for additional evidence in this regard.

Keywords: AHI, Distraction osteogenesis, Obstructive sleep apnea, TMJ ankylosis.

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INTRODUCTION

Maxillomandibular advancement (MMA) surgery is a recommended treatment modality for the management of obstructive sleep apnea (OSA), especially in patients not amenable to conservative, continuous positive airway pressure (CPAP) and oral appliance therapy. Systematic reviews have indicated that this procedure significantly reduces apnea-hypopnea index (AHI) with minimal morbid effects by physically expanding the airway three-dimensionally and preventing its collapse during sleep.^{1,2} A long-term study by Li et al. suggests that the success rate with MMA is greater than 90%.³ The efficacy of MMA is comparable to the gold standard treatment for the condition, that is, CPAP.⁴

Though highly effective, treatment of OSA with MMA by conventional orthognathic surgery is associated with several pitfalls, such as the possibility of limited advancement and limited improvement in the airway; need of subsequent graft and donor site surgery; potentially high relapse³⁻⁶; higher incidence of surgical complications, such as neurosensory deficit^{7,8}; and limited application in syndromic patients, infants, and children due to limited bone stock.^{9,10}

Distraction osteogenesis (DO) has emerged as a viable alternative to conventional orthognathic surgery for the management of maxillomandibular skeletal deformities.¹⁰ Studies have shown that DO allows for greater skeletal expansion with lesser chances of postsurgical relapse and complications.^{10,11} Mandibular corpus distraction osteogenesis (MCDO) is a highly effective procedure for the treatment of OSA secondary to retrognathic

^{1,3}Department of Orthodontics and Dentofacial Orthopedics, Armed Forces Medical College, Pune, Maharashtra, India

^{2,4}Department of Oral and Maxillofacial Surgery, Armed Forces Medical College, Pune, Maharashtra, India

Corresponding Author: SS Agarwal, Department of Orthodontics and Dentofacial Orthopedics, Armed Forces Medical College, Pune, Maharashtra, India, e-mail: docshivagarwal@rediffmail.com

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mandible causing airway constriction. A systematic review has concluded a success rate of 100% and cure rate of 82 to 100% with MCDO in alleviating OSA with marked improvement in AHI and oxygen saturation.⁹ In infants and syndromic cases with midface hypoplasia or mandibular micrognathia, DO may positively affect physical growth and weight gain.¹²

The retrognathic mandible in patients with unilateral or bilateral temporomandibular joint (TMJ) ankylosis may lead to progressive facial asymmetry and development of OSA along with associated symptoms, such as excessive daytime sleepiness, loud snoring,

fatigue, irritability, witnessed apneas, etc. DO is a viable modality in terms of simultaneously improving facial esthetics and possibly curing OSA in these patients.¹³ There is limited literature that highlights the efficacy of DO for patients with severe OSA secondary to TMJ ankylosis, and, therefore, the present study was undertaken for further evidence in this aspect.

Aim

To evaluate the efficacy of MCDO in the management of OSA secondary to TMJ ankylosis.

Objectives

- To evaluate change in skeletal and airway parameters, AHI and Epworth sleepiness scale (ESS) scores following DO.
- To quantify the change in AHI per mm mandibular distraction with DO.

MATERIALS AND METHODS

Study Design

Cross-sectional study.

Study Setting

The patient records available at the archives of the Department of Orthodontics and Dentofacial Orthopedics and Department of Oral and Maxillofacial Surgery of a postgraduate government teaching institution were utilized for the study. Approval from the Institutional Ethical Committee was obtained before the start of the study.

Participants

The treatment records of patients comprehensively managed for severe OSA (AHI score >30) secondary to TMJ ankylosis between 2015 and 2019 with MCDO were selected for the study. All selected patients had undergone an overnight type 1 polysomnography (PSG) at the institute for the diagnosis of OSA prior to treatment and 1 year after distraction surgery. The lateral cephalogram was also obtained at the same time period using a single machine (model: ADVAPX cephalostat machine, company: Panoramic System, and printer: Fujifilm DRY PIX 7000) by the single operator in a standardized technique. Cases selected have been treated by the same orthopedic surgical team with a standard protocol.

Inclusion criteria

- Patients with TMJ ankylosis
- Skeletally mature patients (cervical vertebrae maturation index stage V or VI)
- Patients with primarily obstructive apneas and hypopneas
- Severe OSA (AHI score ≥ 30 per sleep hour)
- Patients treated with mandibular DO only

Exclusion criteria

- Patients with primarily central apneas and hypopneas.
- Syndromic cases including patients with cleft lip and palate
- Patients treated with conventional orthognathic surgery
- Patients who underwent bi-jaw surgery or maxillary DO

Variables

The objective assessment was done by comparing pretreatment (T0, before DO) and 1-year postsurgery (T1) AHI scores on PSG; and superior airway space (SAS), PAS, hypopharyngeal airway space, and mandibular plane-hyoid bone perpendicular distance (MPH) on lateral cephalogram.

Subjective assessment was done at T0 and T1 by ESS survey. The amount of mandibular distraction achieved was noted from the case sheets of the patients. Change in skeletal parameters was assessed by comparison of sella-nasion-point A (SNA), sella-nasion-point B (SNB), and point A-nasion-point B ANB angles at T0 and T1.

Quantification of change in AHI per mm mandibular distraction was done by the following formula:

Mean Change in AHI Score

Mean Mandibular Distraction (mm)

Fifteen patients meeting the inclusion criteria were selected for the study. The mean age of patients selected was 23.3 ± 5.4 years; the minimum–maximum age range was 15 to 33 years; 7 (46.7%) were males; and 8 (53.3%) were females. The distractors were placed under general anesthesia. The distraction was initiated after 5 days of distractor placement at the rate of 1 mm daily (0.5 mm twice daily). The distractors were removed after a consolidation period of 8 weeks. There were 3 cases with unilateral TMJ ankylosis and 12 cases with bilateral TMJ ankylosis. The range of mandibular distraction was 9–19 mm and a mean of 13.3 ± 3.4 mm. Postsurgical (T1) AHI score less than 50% of pretreatment was considered a surgical success, and the achievement of AHI score <5/hour was rated as a surgical cure.^{1,2}

Bias

Ten lateral cephalograms were retraced after a period of 1 week by the principal investigator (SSA) to ascertain intraobserver bias and also by a senior orthodontist to ascertain interobserver bias.

The data obtained were compiled in MS Excel sheets (Microsoft Excel v16.0, Microsoft Corporation, Redmond, Washington) and subjected to statistical analysis.

STATISTICAL METHODS

The data on categorical variables are shown as *n* (% of cases), and data on continuous variables are presented as mean and standard deviation (SD). The pairwise statistical comparisons were done using a paired t-test for each parameter studied. The underlying normality assumption was tested before subjecting each variable to t-test. All the results are shown in tabular as well as graphical format to visualize the statistically significant difference more clearly.

The *p*-values less than 0.05 were considered to be statistically significant in the study. All the hypotheses were formulated using two-tailed alternatives against each null hypothesis (hypothesis of no difference). The entire data were statistically analyzed using Statistical Package for Social Sciences (SPSS version 21.0, IBM Corporation, USA) for MS Windows.

RESULTS

The intra- and interclass correlation coefficient of over 0.8 for the airway parameters on lateral cephalogram indicated a perfect intra- and interobserver agreement.

The mean AHI score at T0 and T1 was 41.71 ± 12.69 and 8.11 ± 3.25 , respectively. The mean AHI score reduction was 33.61 ± 10.56 (*p*-value < 0.001). The mean percentage reduction in AHI score at T1 was 80.39%. The mean AHI score reduction with per mm distraction was 2.59 ± 0.52 (Table 1; Fig. 1).

The mean ESS score at T0 and T1 was 18.60 ± 2.5 and 4.67 ± 1.49 , respectively. The mean decrease in ESS score was 13.93 ± 2.15 (*p*-value < 0.001), and the mean percentage decrease at T1 compared to T0 was 75.05% (Table 2; Fig. 2).

The mean SNA at T0 and T1 was $80.07^\circ \pm 2.31^\circ$ and $79.8^\circ \pm 2.15^\circ$, respectively, with a nonsignificant increase of $0.27^\circ \pm 0.59^\circ$ at T1 (0.32%, p -value > 0.05). The mean SNB at T0 and T1 was $67.8^\circ \pm 5.82^\circ$ and $76.4^\circ \pm 3.5$,

respectively, with an increase of $8.6^\circ \pm 3.42^\circ$ (13.15%, p -value < 0.001). The mean ANB at T0 and T1 was $11.6^\circ \pm 3.68^\circ$ and $3.4^\circ \pm 2.29$, respectively, with a decrease of $8.2^\circ \pm 2.21^\circ$ (72.89%, p -value < 0.001) (Table 3; Fig. 3).

Table 1: Mean pre- and postsurgical AHI scores

	AHI score	
	Mean	SD
Pretreatment (T0)	41.71	12.69
Postsurgical (T1)	8.11	3.25
Reduction (pre-post)	33.61	10.56
Reduction (pre-post) (%)	80.39	
Reduction per mm distraction	2.59	0.52
p -value (pre versus post)	0.001*	

p -value by paired t -test. p -value < 0.05 was considered to be statistically significant. * p -value < 0.001

Table 2: Mean pre- and postsurgical ESS scores

	ESS score	
	Mean	SD
Pretreatment (T0)	18.60	2.50
Postsurgical (T1)	4.67	1.49
Improvement (pre-post)	13.93	2.15
Improvement (pre-post) (%)	75.05	
p -value (pre versus post)	0.001*	

p -value by paired t -test. p -value < 0.05 was considered to be statistically significant. * p -value < 0.001

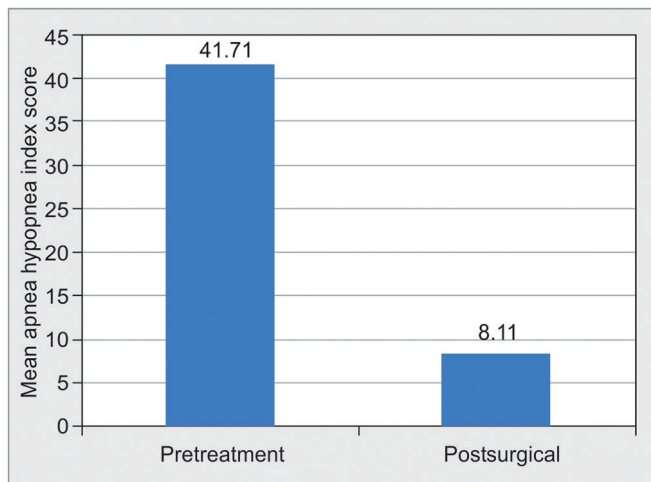


Fig. 1: Mean pre- and postsurgical AHI scores

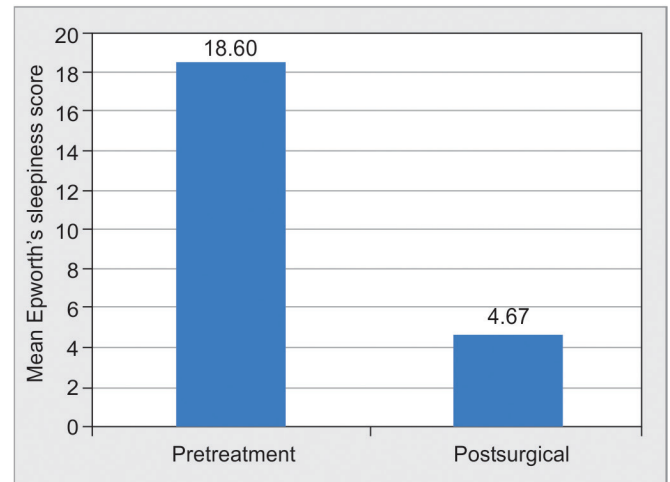


Fig. 2: Mean pre- and postsurgical ESS scores

Table 3: Mean pre- and postsurgical change in skeletal parameters

		Mean	SD
Sella-nasion-point A (SNA) angle	Pretreatment	80.07°	2.31°
	Postsurgical	79.80°	2.15°
	Improvement (pre-post)	0.27°	0.59°
	Improvement (pre-post) (%)	0.32	
	p -value (pre versus post)	0.104 ^{NS}	
Sella-nasion-point B (SNB) angle	Pretreatment	67.80°	5.82°
	Postsurgical	76.40°	3.50°
	Improvement (pre-post)	-8.60°	3.42°
	Improvement (pre-post) (%)	-13.15	
	p -value (pre versus post)	0.001*	
Point A-nasion-point B (ANB) angle	Pretreatment	11.60°	3.68°
	Postsurgical	3.40°	2.29°
	Improvement (pre-post)	8.20°	2.21°
	Improvement (pre-post) (%)	72.89	
	p -value (pre versus post)	0.001*	

p -value by paired t -test. p -value < 0.05 was considered to be statistically significant; * p -value < 0.001; NS: statistically nonsignificant

The mean PAS at T0 and T1 was 4.00 ± 1.46 mm and 10.00 ± 1.25 mm, respectively, with an increase of 6.00 ± 1.07 mm (182.44% increase, p -value < 0.001). The mean SAS at T0 and T1 was 5.00 ± 1.81 mm and 11.93 ± 1.22 mm, respectively, with an increase of 6.93 ± 2.15 mm (179.98% increase, p -value < 0.001). The mean MAS at T0 and T1 was 10.60 ± 3.85 mm and 14.13 ± 1.65 mm, respectively, with an increase of 3.53 ± 3.24 mm (60.76% increase, p -value < 0.001). The mean MPH at T0 and T1 was 25.73 ± 2.05 mm and 16.6 ± 2.03 mm, respectively, with a decrease of 9.13 ± 1.81 mm (35.45% decrease, p -value < 0.001) (Table 4).

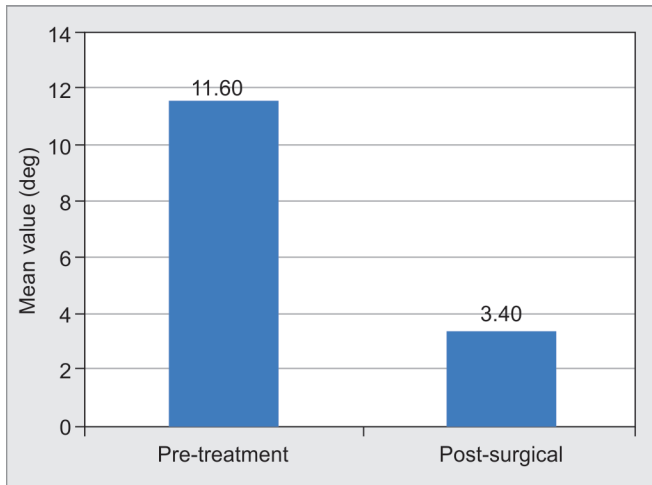


Fig. 3: Mean pre- and postsurgical point A-nasion-point B (ANB) angle

DISCUSSION

TMJ ankylosis is the fusion of the condylar head with glenoid fossa by fibrotic or bony tissues. The TMJ ankylosis in a growing patient will result in mandibular micrognathia and facial asymmetry affecting esthetics, speech, mastication, oral hygiene maintenance, and may cause life-threatening airway compromise. The treatment goals for such patients are to restore form and function, reduce asymmetry and improve facial aesthetics, and achieve a stable occlusion and optimal joint movements postsurgically along with improvement in the airway.^{14,15}

The patients selected for the present study had mandibular retrognathia (mean ANB value of $11.6^\circ \pm 3.68^\circ$) secondary to TMJ ankylosis associated with severe OSA, confirmed by higher AHI scores on PSG. Selected patients were managed with MCDO to achieve the desired goals, including improvement in airway parameters and sleep-related symptoms.

The mean AHI score at T1 (1-year postsurgery) was 8.11 ± 3.25 , which showed 80.39% reduction from the pretreatment values. Postsurgical AHI score showed >50% reduction in all cases, indicating 100% treatment success. At T1, the AHI score in 12 patients was less than 5, which indicated a cure rate of 80%. These findings are similar to other studies in the literature.^{10,15-18} Wang et al.¹⁹ in their observed a complete cure in 23 out of 28 cases studied, whereas 5 patients improved to mild OSA. The mean reduction in AHI score was quantified in the present study as 2.59 ± 0.52 /mm distraction. The authors could not find enough literature in this aspect, and further studies are indicated to validate these findings.

The mean ESS score at T1 (1-year postsurgery) was 4.67 ± 1.49 , which showed a 75.05% reduction indicating a marked improvement

Table 4: Mean pre- and postsurgical airway parameters

		Mean	SD
Posterior airway space (PAS) (mm)	Pretreatment	4.00	1.46
	Postsurgical	10.00	1.25
	Improvement (pre-post)	-6.00	1.07
	Improvement (pre-post) (%)	-182.44%	
	p -value (pre versus post)	0.001*	
Superior airway space (SAS) (mm)	Pretreatment	5.00	1.81
	Postsurgical	11.93	1.22
	Improvement (pre-post)	-6.93	2.15
	Improvement (pre-post) (%)	-179.98%	
	p -value (pre versus post)	0.001*	
Minimum airway space (mm)	Pretreatment	10.60	3.85
	Postsurgical	14.13	1.65
	Improvement (pre-post)	-3.53	3.24
	Improvement (pre-post) (%)	-60.76%	
	p -value (pre versus post)	0.001*	
Mandibular plane-hyoid (MPH) (mm)	Pretreatment	25.73	2.05
	Postsurgical	1.60	2.03
	Improvement (pre-post)	9.13	1.81
	Improvement (pre-post) (%)	35.45%	
	p -value (pre versus post)	0.001*	

p -value by paired t -test; p -value <0.05 was considered to be statistically significant; * p -value <0.001; NS, statistically nonsignificant

in sleep-related symptoms of the patient, such as excessive snoring, excessive daytime sleepiness, and tendency to sleep in unusual situations, such as watching television or while driving. These findings are similar to another study on moderate to severe OSA cases, which reported improvement of ESS score from 10.25 to 2.25.¹⁸ The mean pretreatment ESS score was higher in the present study (18.60 ± 2.5) as only severe OSA cases were studied.

In the present study, the increase in length of the mandible (distraction) was shown by an increase in SNB from $67.8^\circ \pm 5.82^\circ$ pretreatment to $76.4^\circ \pm 3.5^\circ$ at T1, that is, 13.15% increase; and decrease in ANB from $11.6^\circ \pm 3.68^\circ$ to $3.4^\circ \pm 2.29^\circ$, that is, 72.89% decrease. This indicated a marked improvement in the profile of the patient and optimization of skeletal bases, which is similar to the existing literature.^{19,20} Wang et al.¹⁹ in their study observed a mean increase in SNB angle from 66° to 75° , which is similar to our study wherein SNB increased from $67.8^\circ \pm 5.82^\circ$ to $76.4^\circ \pm 3.5^\circ$. However, the quantum of skeletal change depends upon the amount of mandibular advancement done in various studies.

The mean airway parameters studied, that is, PAS, SAS, MAS, and MPH, showed statistically significant marked improvement at T1 (182.44%, 179.98%, 60.76%, and 35.45% increase, respectively). The overall improvement in the airway in the present study is similar to that observed by a systematic review.¹⁰ These changes are similar to a study by Mohamed²¹ wherein computed tomography (CT) scans were used to study postdistraction airway changes that were found to be 141% of pretreatment. The subjects of OSA secondary to Pierre Robin sequence were studied in the above study, whereas in ours, the subjects had TMJ ankylosis as the causative factor of OSA and lateral cephalogram was used instead of CT. Studies have shown that the accuracy of lateral cephalograms is comparable to that of CT in diagnosing airway problems.²² Wang et al.¹⁹ in their study observed an increase in postdistraction PAS from 4.6 to 12.5 mm (172% increase), which is similar to the present study (182.44%). The airway increase in the present study is lesser as compared to another study,²³ which observed 209% improvement in PAS. The postsurgical improvement in the airway in the present study is higher as compared to another study,²⁴ which observed 71.9% increase in a 3D CT-based study in children.

CONCLUSION

The results of this study indicate that MCDO is an effective modality for the treatment of severe OSA secondary to TMJ ankylosis with a success rate of 100% and a cure rate of 80%. The mean reduction in AHI score per mm distraction was observed as 2.59 ± 0.52 mm. Since there is the scantiness of the literature in this aspect, the authors recommend further studies for additional evidence in this regard.

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