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Distraction osteogenesis in the management of mandibular hypoplasia secondary to temporomandibular joint ankylosis. Long term follow up

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ABSTRACT

Purpose: The aim of this retrospective study was to evaluate the short-term and long-term skeletal and soft-tissue stability after MDO with or without genioplasty, as well as the stability of the achieved maximum inter-incisal opening (MIO) in patients with mandibular hypoplasia secondary to TMJ ankylosis.

Patients and methods: Twenty patients with mandibular hypoplasia secondary to TMJ ankylosis were managed by a two-stage surgical protocol, gap arthroplasty as the first stage, followed by MDO. The patients were analyzed for skeletal and soft-tissue stability as well as the maintenance of the achieved MIO. Lateral cephalograms were evaluated at four time intervals: pre-distraction (T1), after a consolidation period with or without genioplasty (T2), after one year following consolidation (T3), and at the longest follow-up (T4). Statistical analyses compared the skeletal and soft-tissue changes at different intervals in every group.

Results: All the ankylosed joints except three were treated with gap arthroplasty without costochondral graft. The MIO was increased from 8.2 ± 2.1 mm preoperatively to 40.2 ± 1.7 mm postoperatively. After the consolidation period, MIO decreased to 23 ± 6.5 mm. The patients were instructed to restart active physiotherapy after removal of the distractors to regain the pre-distraction MIO, which was maintained during the short-term follow-up. The mean follow-up period was 8.5 ± 1.5 years. At the end of the follow-up, two patients showed recurrence of ankylosis. Cephalometric analysis revealed great improvements in the hard- and soft-tissue structures after MDO with or without genioplasty. Several significant long-term relapses could be observed in all groups; however, they did not reach their preoperative values.

Conclusion: TMJ ankylosis leads to severe, multidirectional mandibular hypoplasia, which is significantly corrected with the MDO. The MDO provides a stable short-term improvement in the facial esthetics at the first postoperative year, but a significant relapse occurs during the long term follow-up. Nevertheless, a satisfied facial esthetic is maintained for up to seven to 12 years postoperatively. During the activation period, the MDO minimizes the gained MIO after release of ankylosis, but the MIO is successfully restored with physiotherapy.

1. Introduction

Temporomandibular joint (TMJ) ankylosis is one of the most serious and disabling conditions that challenge oral and maxillofacial surgeons. The challenge not only lies with the restoration of the joint function but also with the management of the associated complex secondary deformities (Troulis et al., 2008). Different surgical techniques have been described for the treatment of TMJ ankylosis, including gap arthroplasty, inter-positional arthroplasty, and costochondral grafts reconstruction, resulting in significantly improved maximum inter-incisal opening (MIO) and favorable jaw function. Even after successful surgical release of the ankylotic mass, the mandibular deformity may become aggravated because of the loss of the ramus height. Recently, distraction osteogenesis (DO) is gaining popularity in the management of mandibular

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hypoplasia subsequent to TMJ ankylosis (Mehrotra et al., 2016). Postoperative stability is considered one of the main advantages of DO. Gradual stretching of the surrounding soft-tissue matrix, less periosteal stripping, and positioning of the osteotomy site distal to the pterygomasseteric sling have been considered beneficial to the stability of the technique (Schreuder et al., 2007).

DO relapse is defined as the gradual recurrence of the deformity for which distraction was performed. The magnitude of mandibular advancement (Serafin et al., 2007) and the large mandibular plane angle may influence the stability of mandibular distraction osteogenesis (MDO) (Joss et al., 2013a). In addition, the regenerate in MDO is subjected to many external forces during consolidation, including those from the surrounding soft-tissue matrix. These forces can alter the position, size, and shape of the post-distracted bone (Al-Daghreer et al., 2008). Relapse in MDO has been reported to be minimal, because of the gradual lengthening of the surrounding soft-tissue matrix (Strijen Van et al., 2004; Ko et al., 2004; Baas et al., 2015). However, many authors reported significant relapse during the follow-up period (Sadakah et al., 2006; Baas et al., 2015; Peacock et al., 2018). These controversies between results were present because different methods of evaluation and different timings have been used during follow-up.

Although the DO technique has been in use for more than 25 years, no definitive conclusions have been reached on the long-term stability of the distracted hypoplastic mandible secondary to TMJ ankylosis. The purpose of this retrospective cephalometric study was to evaluate the short-term and long-term skeletal and soft-tissue stability after MDO with or without genioplasty, as well as the stability of the achieved MIO in patients with mandibular hypoplasia secondary to TMJ ankylosis.

2. Patients and methods

2.1. Study design and population

The initial study comprised 28 adult patients who had mandibular hypoplasia secondary to TMJ ankylosis. They were selected from the outpatient clinic of the Department of Oral and Maxillofacial Surgery, Faculty of Dental Medicine for Girls, Al Azhar University, between June 2004 and May 2009. A two-stage surgical protocol was followed; the TMJ ankylosis was released in the first stage through gap arthroplasty, and then the facial deformity was corrected in the second stage by MDO. Patients who fulfilled the following inclusion criteria were included in the study: (1) a long-standing TMJ ankylosis with secondary mandibular hypoplasia, (2) non-growing adult patients, (3) mandibular advancements >15 mm, (4) at least 12 months should have elapsed between the release of the ankylosis and the MDO, and (5) a follow-up period of one to seven years should have elapsed after the MDO for short-term and long-term follow-up, respectively. Patients were excluded if they were syndromic or medically compromised. Written informed consent was obtained from all patients, and the local ethics review committee of the Faculty of Dental Medicine for Girls at Al Azhar University approved the study.

In 2016, all patients were contacted by telephone and asked to participate in a long-term clinical and cephalometric follow-up examination. Twenty (71.4%) out of 28 patients accepted the invitation, three patients (10.7%) did not respond to recall, two patients (7.15%) wanted to participate but were busy during the time of data collection, two patients (7.15%) moved out of the country, and the last patient (3.6%) was pregnant. The 20 patients were redistributed into three groups; group 1 included seven (35%) patients who refused genioplasty after MDO; group 2 included five (25%) cases with unilateral mandibular deficiency; and the remaining eight (40%) subjects, who had a bilateral mandibular deficiency, were included in group 3. All the patients in groups 2 and 3 underwent MDO followed by advancement genioplasty at the end of the consolidation period.

2.2. Study variables

The predictable variables were the patients' demographic and anatomic variables. The demographic variables included the age of the patients at the time of the MDO operation; their gender; the etiology of the deformity, the amount of mandibular advancement, and the duration of activation; and the follow-up periods. The anatomic variables were the amount of mandibular deficiency that was measured in vertical and horizontal dimensions for both soft and hard tissues, and the pre-distraction MIO. The primary outcome variable was the analysis of the hard- and soft-tissue changes occurring after MDO with or without genioplasty in both vertical and horizontal dimensions. The secondary outcome variables were the hard- and soft-tissue stability during the short-term and long-term follow-up periods as well as the stability of the MIO.

2.3. Treatment protocol

2.3.1. Release of TMJ ankylosis

At the first stage of the patient's treatment, an orthopantomogram (OPG) and computed tomography (CT) were requested to detect the extent of the ankylosis. In addition, an airway assessment was performed by polysomnography (PSG) that included the apnea–hypopnea index (AHI) and average oxygen saturation to detect the degree of obstructive sleep apnea (OSA) in all patients. All but three patients underwent gap arthroplasty without costochondral graft. For prevention of re-ankylosis, physiotherapy by using a mouth gag was done, and the patients were instructed to continue physiotherapy for 12 months.

2.4. MDO protocol

2.4.1. Preoperative assessment (T1)

The diagnosis of mandibular hypoplasia and planning of distraction vector were made by clinical and radiographic examinations: cephalograms, OPG, and CT. The cephalograms (lateral and posteroanterior) were performed to determine the amount of mandibular deficiency. In bilateral cases, the oblique vector was chosen to lengthen the mandibular body and ramus. To achieve that, the angle between the long axis of the distractors and the maxillary occlusal plane ranged from 30° to 40°. To correct the facial asymmetry and maxillary occlusal cant in patients with unilateral mandibular deficiency, a simultaneous maxillomandibular distraction was performed. In those cases, a vertical vector was selected, in which the angle between the long axis of the distractor and the maxillary occlusal plane ranged from 45° to 90°. In all cases, the osteotomy was designed to be perpendicular to the distraction vector.

2.5. MDO surgical procedures

All the surgical procedures were performed under general anesthesia, by the same oral and maxillofacial team. Extraoral multidirectional mandibular distractors (Stryker-Leibinger Multiguide, Howmedica Leibinger GmbH and Co. KG, Freiburg, Germany) were used to lengthen and adjust the mandible in three directions: linear, angular, and transverse. Through an intraoral approach, at the line of the planned osteotomy, a buccal corticotomy and superior and inferior osteotomies with a surgical reciprocating saw were performed under sterile saline irrigation. The lingual cortex and medullary bones were disrupted by a greenstick

maneuver with an osteotome. This was to prevent injury of the inferior alveolar nerve (IAN). The extraoral distractor was applied and fixed in position, using four bicortical, stainless steel, self-drilling pins (2 mm in diameter and 9 mm in length) through an extraoral trocar.

In the unilateral cases, when simultaneous maxillomandibular distraction was planned, a complete Le Fort I osteotomy was performed and down-fractured. The maxilla was mobilized, and a transosseous wire was inserted into the contralateral zygomatic buttress to act as a fulcrum, and then the mandibular osteotomy site was done as before. The maxillomandibular fixation was performed to allow simultaneous movement of the maxilla with the mandible during the distraction process as a single unit. Before closure of the surgical wounds, the distractor was activated to confirm the proper functioning and completion of the osteotomy. After that, the distractor was returned to its original position. The MDO was started after a five-day latency period and performed at the rate of 1 mm/day. Overcorrection was done in all cases to overcome the expected post-distraction relapse.

In bilateral cases, after the desired mandibular lengthening was achieved, the angle of the distractors was closed, and anterior elastics were applied to close the anterior open bite (AOB), which was developed during the distraction process. On completion of the molding procedure, the consolidation period started and lasted for 12 weeks (until osteogenesis was confirmed in the distraction gap by radiograph). At the end of the consolidation period, the distractors were removed under local anesthesia in the seven patients (group 1) who refused genioplasty. Under general anesthesia, removal of the distractors was performed, followed by advancement genioplasty for patients who accepted this procedure (groups 2 and 3). Genioplasty was performed through mandibular vestibular incision. An osteotomy was created to separate the chin to allow its anterior advancement to the preoperatively planned distance. The genial segment was then stabilized by using interosseous wires.

2.6. Postoperative assessment

All patients were assessed clinically and radiographically at three postoperative time intervals: (1) T2, when the assessment was done at the end of the consolidation period for patients in group 1, in groups 2 and 3, after genioplasty; (2) T3, recorded one year after the consolidation period; and (3) T4, performed at least seven years after the consolidation period. By using lateral cephalometric analysis, the short-term and long-term hard- and soft-tissue changes were evaluated throughout the study's follow-up period. The amount of change was recorded by calculating the difference between the measurement of each cephalometric point at a certain time interval and its previous one as follows: the measurements of T2–T1 were used to assess the results of MDO with or without genioplasty. Those of T3–T2 were used to assess the short-term results, whereas those of T4–T3 determined the long-term results. In addition, the overall changes were calculated by comparing the long-term changes and the pretreatment cephalometric values (T4–T1). By using the posteroanterior (PA) cephalometric analysis, the degree of facial asymmetry was also recorded.

The MIO was clinically recorded. The distance between the incisal edges of the upper and lower central incisors was measured by using a caliper. The patients' satisfaction was also evaluated through a questionnaire in a multiple-choice format that included nontechnical terminology for patients of different educational backgrounds. The questionnaire included questions about the improvement in facial esthetics, mandibular function, and feedback from other observers. The level of satisfaction was graded from 1 to 5 as follows: 1 (not satisfied), 2 (slightly satisfied), 3 (moderately satisfied), 4 (satisfied), and 5 (completely satisfied). Furthermore, the intraoperative, short-term, and long-term postoperative complications were documented. Evaluated complications included paresthesia, pain during device activation, loosening of pins, postoperative infection, hardware failure, development of AOB, soft-tissue scarring, tooth injuries, recurrence of OSA, and re-ankylosis. These complications were subdivided according to their severity into minor and major complications.

2.7. Methodology of the cephalometric analysis

The patients were assessed for short-term and long-term hard- and soft-tissues changes by using lateral cephalometry. Tracings were made on acetate paper, and the cephalometric points and lines were identified according to Burstone et al. (1978). The definitions of the reference lines and cephalometric landmarks are listed in Fig. 1 and Table 1. To assess the positional changes of the mandible, horizontal and vertical reference lines were used. The horizontal line (HL) was drawn 7° from the Sella–Nasion (SN, anterior cranial base) line. A line perpendicular to the HL was then drawn through the nasion as a vertical reference line (VL). The linear and angular measurements are listed in Table 2. To evaluate the direction of displacement in the horizontal dimension, the

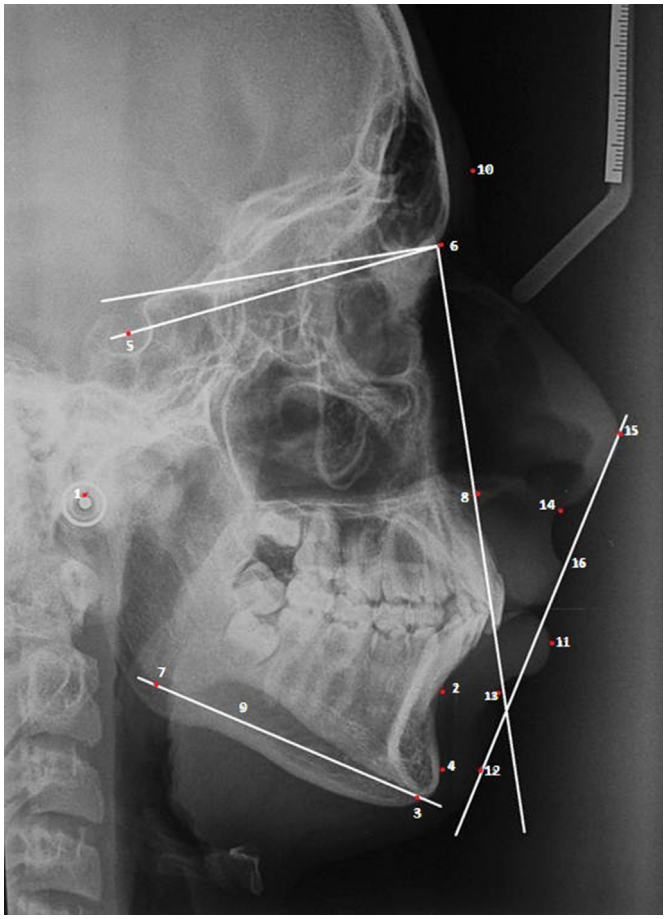

Fig. 1. Reference lines and cephalometric points used in this study. The hard- and soft-tissue landmarks are defined in Table 1. 1. Articular (Ar), 2. B point (B), 3. Menton (Me), 4. Pogonion (Pg), 5. Sella (S), 6. Nasion (N), 7. Gonion (Go), 8. Anterior nasal spine (ANS), 9. Mandibular plane (MP), 10. Glabella (G), 11. Labrale inferious (Li), 12. Soft-tissue pogonion (Pgs), 13. Soft-tissue B point (Bs), 14. Subnasale (Sn), 15. Pronasal (Prn), 16. Esthetic line (E-line).

Table 1
Cephalometric landmarks used in this study.

Hard-tissue landmarks	
Ar	Articulare: the intersection of the posterior ramus and inferior cranial base
B point	Deepest point on the anterior curvature of the mandibular alveolar process
Me	Menton: the lowermost point on the symphysis of the mandible
Pg	Pogonion: the most anterior point on bony chin contour
S	Sella: Centre of sella turcica
N	Nasion: most anterior point of the frontonasal suture
Go	Gonion: midpoint of the angle of the mandible
ANS	Anterior nasal spine: The most anterior extremity of the intermaxillary suture
MP	Mandibular plane passing through gonion to menton
Soft-tissue landmarks	
G	Glabella: the most anterior point of the soft-tissue forehead
Li	Labrale inferious: the most anterior point of the lower lip
Pgs	Soft-tissue pogonion: the most anterior point of soft-tissue chin contour
Bs	A point of greatest concavity in the midline of the lower lip between Li and Pgs
Sn	Subnasale: point at which columella merges with the upper lip
Prn	Pronasal: soft-tissue point on tip of the nose
E line	Esthetic line, a line connecting Prn and Pgs

positive values represented a forward movement of the point, whereas the negative values represented the relapse that occurred in a posterior direction (i.e., toward the preoperative position). Similarly, in the vertical plane, the positive and negative values were assigned to indicate downward and upward displacement, respectively. The measurements were taken twice by the same examiner, one month apart, and the mean outcome of both measurements represented the final results.

2.8. Statistical analyses

The collected data was recorded, tabulated, and analyzed statistically. The data was analyzed with Microsoft Office XP (Excel) and SPSS version 15.00 software (SPSS Inc., Chicago, IL, USA). Age of the patients, duration of the distraction and the follow-up periods, the value of cephalometric changes, and the MIO were described using the mean and standard deviation (SD). A paired sample *t*-test was used to identify significant changes in the cephalometric variables at the four time points, and *p*-values below 0.05 were considered statistically significant. The relationships among cephalometric variables, amount of advancement, age, and gender of the patients were analyzed with the Pearson correlation coefficient.

Table 2
Linear and angular measurements used in this study.

Linear and Angular skeletal measurements	
Horizontal	
- B–VL	To measure the anterior position of the mandible
- Pg–VL	To indicate bony chin prominence
- Go–Pg	Length of the corpus of the mandible
Vertical	
- Ar–Go	Mandibular ramus height
- ANS–Me	Anterior lower facial height
- MP–HL°	To measure positional changes of the mandible relative to the cranial base
- Ar–Go–Me°	Gonial angle (Angle between the ramus and the corpus of the mandible)
Linear soft-tissue measurements	
Horizontal	
- Bs–VL	Distance between soft-tissue B point and vertical line
- Pgs–VL	Distance between soft-tissue pogonion and vertical line
- G–Sn–Pgs°	Facial convexity angle
- Li–E line	Lower lip protrusion to esthetic line

The degree of significance was classified as follows: strong correlation ($r > 0.8$), moderate correlation ($r = 0.5–0.8$), and weak correlation ($r < 0.5$).

3. Results

Twenty patients were accepted in this retrospective, long-term study. The patients' population consisted of 14 females (70%) and six males (30%). Their mean age at surgery was 20.5 ± 3.4 years. Of the 20 patients, 12 patients underwent unilateral MDO (60%), and the remaining eight patients underwent bilateral MDO (40%), so that the study included a total of 28 mandibular distraction sides. Seven (35%) of the unilateral cases had mandibular deficiency on the left side, whereas right-side deficiency was recorded in five cases (25%). All the patients had facial deformities secondary to TMJ ankylosis. Nineteen patients gave a history of trauma during early childhood. The main cause of the trauma was falls that were reported in 17 cases (85%) and two cases (15%) reported that the cause was a road traffic accident. Only one case among our patients gave a history of odontogenic infection. The mean preoperative MIO for all the subjects was 8.2 ± 2.1 mm (range 0–15), which increased and maintained a mean of 40.2 ± 1.7 mm (range 30–45) after 12 months postoperatively. The mean duration of active distraction was 46.7 ± 10.7 days (range 30–60). The mean follow-up period was 8.5 ± 1.5 years and ranged from seven to 12 years. The demographic features of the study's patients are summarized in Table 3.

Patients of group 1 (seven cases) still had a retruded chin as documented by cephalometric analysis; nevertheless, they were satisfied with their facial appearance and refused to do advancement genioplasty. In groups 2 and 3 (five and eight patients, respectively), the advancement genioplasty was undertaken as a secondary camouflage procedure (Fig. 2) to provide better esthetic outcomes. During the activation period, the MIO measurements decreased by a mean of 23 ± 6.5 mm. The patients were instructed to restart active physiotherapy after removal of the distractors, and the pre-distraction MIO measurements were regained in all the patients. During the short-term follow-up, the MIO was maintained in all the patients with a mean of 39.7 ± 1.4 mm (range 29–43). At the end of the follow-up, the MIO was 36.05 ± 13 mm (range 10–43), including 18 patients (90%) with values exceeding 31 mm

Table 3
Demographic features of the patients.

	Descriptive analysis
Patients number	20
Group 1	7
Group 2	5
Group 3	8
Mean age at operation (years)	20.5 ± 3.4
Group 1	19.3 ± 3
Group 2	21.2 ± 4.2
Group 3	21.1 ± 3.3
Mean duration of active distraction (range), days	46.7 ± 10.7 (30–60)
Mean duration of follow-up (range), years	8.5 ± 1.5 (7–12)
Gender	
Female/male	14/6
Group 1	5/2
Group 2	4/1
Group 3	5/3
Etiology of the deformity	
Trauma	19
Infection	1
Location of TMJ ankylosis	
Unilateral	12
Right	5
Left	7
Bilateral	8

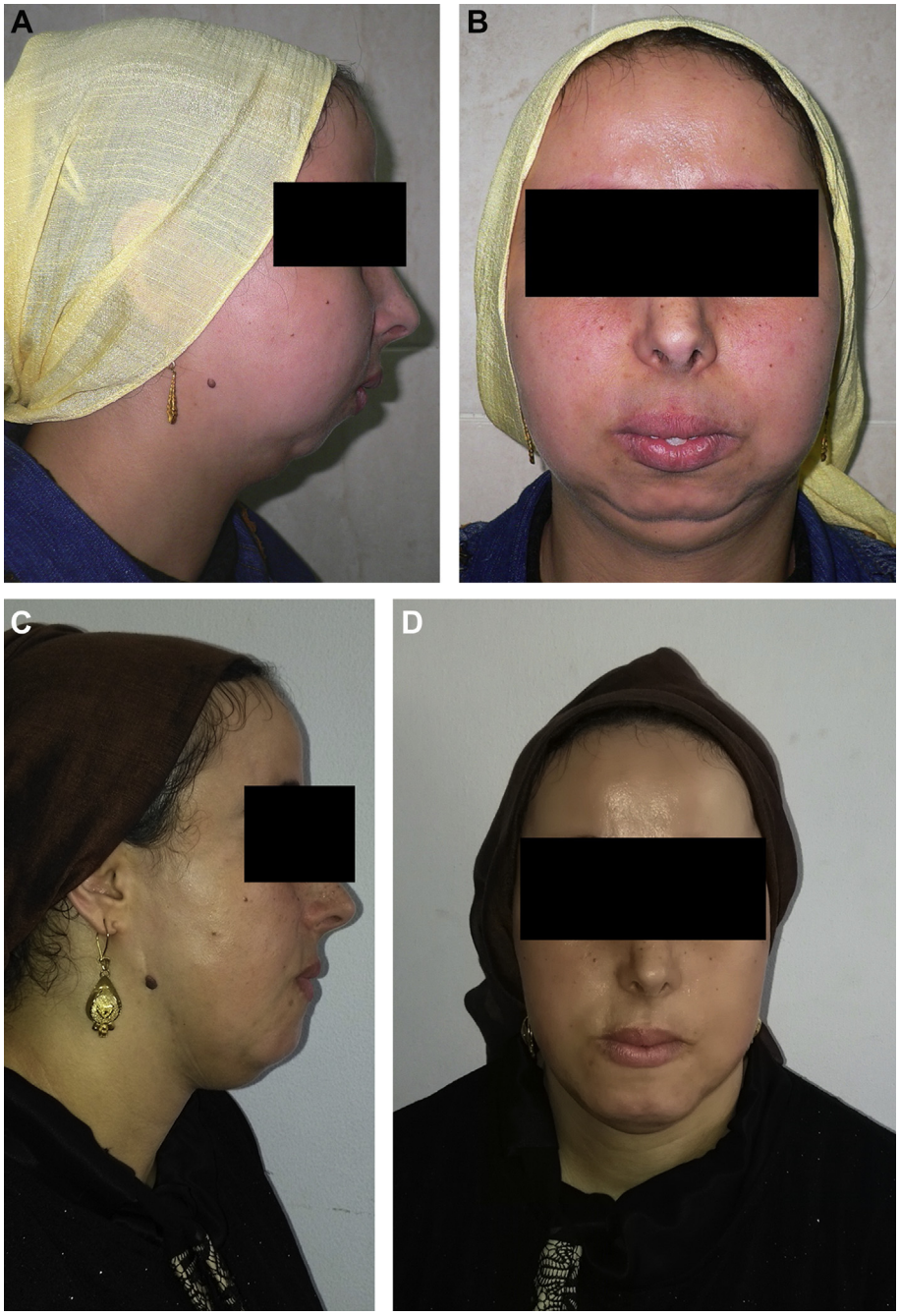

Fig. 2. Clinical extraoral photographs for patient of group 3: (A) lateral and (B) frontal views before mandibular distraction osteogenesis surgery; (C) lateral and (D) frontal views at the longest follow-up period (8 years).

and two patients (10%) showed values below 12 mm because of recurrence of the TMJ ankylosis, which was confirmed by CT.

3.1. Cephalometric analysis

The cephalometric analysis revealed a great improvement in the hard- and soft-tissue structures after MDO in all groups (Fig. 3). The descriptive statistics and statistical comparisons for cephalometric changes at all the follow-up periods are presented in Tables 4–9.

3.1.1. T2–T1 time interval

In all groups, the soft-tissue points (Bs, Pgs) were significantly advanced following the horizontal movements of the corresponding skeletal points (B, Pg). In addition, there was a significant increase in the length of the corpus of the mandible (Go–Pg), ramus height, and lower facial height (ANS–Me). The mandibular plane and facial convexity angles showed a significant reduction in their values. On the other hand, the gonial angle exhibited a significant decrease in groups 1 and 2. There was a significant decrease in the distance between the Li point and the E line which indicated that the Li point advanced anteriorly in all patients (Tables 5, 7 and 9).

3.1.2. T3–T2 time interval (short-term follow-up)

The short-term post-distraction evaluation of group 1 revealed a minimal degree of relapse in both hard and soft tissues, but it was

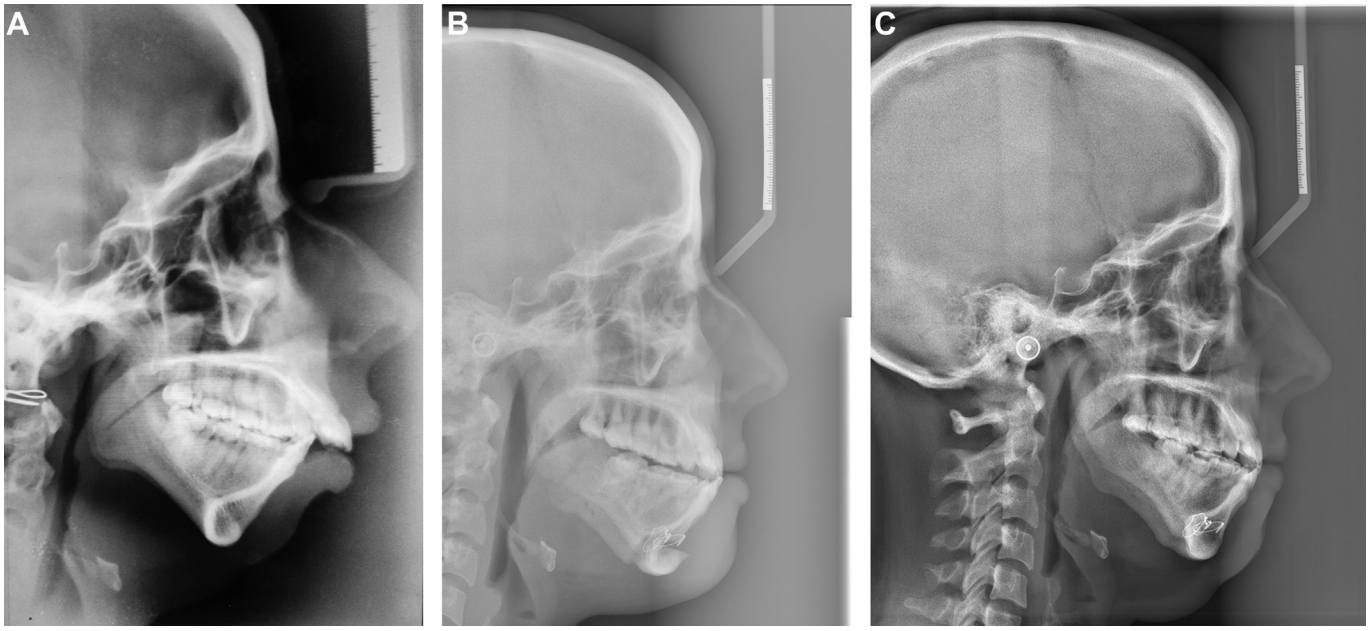

Fig. 3. Lateral cephalometric X-rays of the patient in Fig. 2. (A) pre-distraction, (B) after the consolidation period and advancement genioplasty, and (C) at the longest follow-up period (8-years).

statistically insignificant (Table 5). On the other hand, a significant relapse was observed in group 2 regarding the following parameters: the ramus height (−2.4 ± 1.7), MP–HL° (4.2 ± 2.3), gonial angle (3.4 ± 1.7), Bs–VL (−3 ± 0.7), and facial convexity angle (2.4 ± 0.9) as shown in Table 7. In group 3, the results only showed a significant relapse in the following points: Bs–VL (−1.8 ± 1.5), Pgs–VL (−1.4 ± 1.3), and facial convexity angle (1 ± 1) (Table 9).

3.1.3. T4–T3 time interval (long-term follow-up)

The majority of the cephalometric parameters exhibited a statistically significant relapse at the end of the follow-up in all groups. Nevertheless, the degree of relapse did not reach their preoperative values (Tables 5, 7 and 9).

Table 4
Descriptive statistics for cephalometric variables at all the follow-up periods (group 1).

	T1		T2		T3		T4	
	Mean	SD	Mean	SD	Mean	SD	Mean	SD
Skeletal measurements								
Horizontal								
B–VL (mm)	−19.1	5.5	−13.7	9.8	−14.4	6.3	−16.4	6.4
Pg–VL (mm)	−21.7	5	−16.1	8.5	−17.3	7.6	−18.6	7
GO–Pg (mm)	64.3	9.6	70.6	7.9	69.9	7.8	69.1	7.4
Vertical								
ANS–Me (mm)	59.7	7.5	69.3	4.6	68.7	4.9	66.6	4.9
Ar–GO (mm)	37.6	3.9	57.1	7.8	55.4	7.9	53.3	7.8
MP–HL (°)	39.6	7.6	27.1	6.9	29.7	5.6	33.1	6.4
Ar–GO–Pg (°)	135	7.5	120.7	3.8	122.6	4.7	127.4	5
Soft-tissue measurements								
Horizontal								
Bs–VL (mm)	3.3	3.3	2.7	6	1.3	5.2	0	4.6
Pgs–VL (mm)	−5.1	4.7	2	5.3	0.9	4	−1.1	4.5
Li–E line (mm)	4	1.2	2	1.2	2	1.2	2.4	0.8
G–Sn–Pgs (°)	29.3	5.8	21.4	4.9	22.9	5.2	24.6	5

NOTE. Cephalometric variables are given in degrees and mm.
Abbreviations: T1, pre-distraction; T2, after consolidation period with or without genioplasty, T3, one year after the consolidation period; and T4, at the longest follow-up at least seven years after consolidation. SD, standard deviation. For other abbreviations, see Tables 1 and 2.

3.1.4. T4–T1 time interval

When comparing the long-term results to the pretreatment cephalometric values, significant improvements were observed in all groups (Tables 5, 7 and 9).

3.1.5. Results of the statistical correlations

At the end of the follow-up period (T4–T3), the degree of association between the variable changes during treatment was evaluated. No statistically significant correlations were found between the amount of relapse that occurred in the cephalometric parameters and the gender or age of the patients, and no correlations were found between the degree of relapse of cephalometric variables and the amount of mandibular advancement.

3.2. Relationship between hard- and soft-tissue pogonion

In the horizontal direction, the Pg and Pgs changes throughout the entire observation period (T4–T1) were strongly correlated. In group 1, the ratio between the Pg to Pgs changes was 1:1.3 with a moderate and nonsignificant correlation ($r = 0.65$, $p = 0.11$). In group 2, the Pg to Pgs ratio was 1:1.1, and the correlation of Pgs to Pg was strong and significant ($r = 0.95$, $p = 0.001$). In group 3, the ratio was 1:1 with a strong and significant correlation of Pgs to Pg ($r = 0.9$, $p = 0.003$).

3.3. Relationship between B and Bs points

In all groups, the Pearson's correlation coefficient in the horizontal direction revealed that there was a nonsignificant moderate correlation between B and Bs points ($r = 0.7$, $p = 0.5$), and the ratio between the B to Bs changes was 1:1.2.

3.4. Patients' satisfaction

All the patients stated that the surgery met their expectations, and they were satisfied with their facial esthetics and mandibular function, except that two patients were not satisfied with function because of the recurrence of TMJ ankylosis, but they were satisfied

Table 5
Analysis of mean changes for hard- and soft-tissue measurements (group 1).

	T2–T1			T3–T2			T4–T3			T4–T1		
	Mean	SD	*p*-value	Mean	SD	*p*-value	Mean	SD	*p*-value	Mean	SD	*p*-value
Skeletal measurements												
Horizontal												
B–VL (mm)	5.4	2.6	0.002	−0.71	1.9	0.4	−1.93	1	0.003	2.8	1.4	0.002
Pg–VL (mm)	5.6	4.3	0.014	−1.1	2.2	0.21	−1.4	1.2	0.03	3	2.5	0.02
GO–Pg (mm)	6.3	6.2	0.04	−0.7	1.5	0.25	−0.7	1.1	0.14	4.9	3.9	0.02
Vertical												
ANS–Me (mm)	9	4	0.0009	−0.3	0.8	0.4	−2.3	1	0.0007	6.4	4.5	0.009
Ar–GO (mm)	19.6	6.3	0.0002	−1.7	3	0.2	−2.14	0.9	0.0007	15.7	6.4	0.0006
MP–HL (°)	−12.4	5	0.0006	2.6	3.1	0.07	3.4	2.7	0.02	−6.4	3.2	0.002
Ar–GO–Pg (°)	−15	4.5	0.0001	1.9	2.9	0.13	4.9	2.6	0.003	−8.3	3.6	0.0009
Soft-tissue measurements												
Horizontal												
Bs–VL (mm)	6	3	0.002	1.43	2.1	0.13	1.3	1	0.011	3.3	2	0.005
Pgs–VL (mm)	7.1	4	0.003	−1.1	2	0.2	−2	0.8	0.0006	4	2.7	0.008
Li–E line (mm)	−2	0.8	0.0006	0	0	1	0.4.	0.5	0.08	1.6	0.8	0.002
G–Sn–Pgs (°)	−7.9	2.8	0.0003	1.4	1.8	0.08	1.7	0.8	0.0009	−4.7	1.6	0.0002

*Significant comparison, $p \leq 0.05$.

Table 6
Descriptive statistics for cephalometric variables at all the follow-up periods (group 2).

	T1		T2		T3		T4	
	Mean	SD	Mean	SD	Mean	SD	Mean	SD
Skeletal measurements								
Horizontal								
B–VL (mm)	−29.2	3.3	−21.6	2.7	−21.8	2.6	−24	1.6
Pg–VL (mm)	−30	4.5	−19.4	2.4	−20	3	−24.2	2.6
GO–Pg (mm)	58.6	2.7	72.2	1.5	70.8	1	68.2	2.2
Vertical								
ANS–Me (mm)	58.8	3.4	69	3.5	67.6	4	65.4	4.4
Ar–GO (mm)	39	2.2	57	6.5	54.6	1.1	51.8	1.3
MP–HL (°)	45	3.3	34	3.3	38.2	4.3	39.4	3.4
Ar–GO–Pg (°)	134.8	7.7	122.2	2.9	125.6	2.6	128.6	3.4
Soft-tissue measurements								
Horizontal								
Bs–VL (mm)	−13.4	5.5	−1.6	7.2	−4.6	7.2	−7	5.1
Pgs–VL (mm)	−10.4	6.7	−0.2	4	−0.6	3.8	−3.8	3.1
Li–E line (mm)	5.8	1.6	1.4	1.3	1.4	1.3	2.4	1.8
G–Sn–Pgs (°)	32.4	3.4	18.8	3.6	20.6	3.4	23.4	2.6

NOTE. Cephalometric variables are given in degrees and mm.

Table 8
Descriptive statistics for cephalometric variables at all the follow-up periods (group 3).

	T1		T2		T3		T4	
	Mean	SD	Mean	SD	Mean	SD	Mean	SD
Skeletal measurements								
Horizontal								
B–VL (mm)	−35.4	8.2	−23.4	4.6	−24	5	−26.3	5
Pg–VL	−44.1	9.9	−22.6	7.9	−23.8	7.6	−28.5	8.3
GO–Pg (mm)	60.8	8.8	72.8	6	72.5	5.7	70.4	6
Vertical								
ANS–Me (mm)	61.6	7.6	70.9	4.3	70.3	4.2	68.5	4.1
Ar–GO (mm)	43.1	6.9	54.1	6	52.8	5.5	50.8	5.3
MP–HL (°)	46	8.6	37.6	7	38.9	6.6	41	6.6
Ar–GO–Pg (°)	120.8	8.8	124.5	4.7	125.5	4.4	126.1	4.3
Soft-tissue measurements								
Horizontal								
Bs–VL (mm)	−17.3	13.5	−2.5	10.8	−4.3	10.6	−6.6	10.5
Pgs–VL (mm)	−26.3	14.8	−6.3	11.2	−7.6	11.4	−10.3	12.3
Li–E line (mm)	10.3	4.6	2.6	4.2	3	4.5	4.6	4.5
G–Sn–Pgs (°)	37.1	6.2	19.1	5	20.1	4.7	22.9	5.5

NOTE. Cephalometric variables are given in degrees and mm.

with their facial esthetics (Table 10). No major complications were encountered intraoperatively in this study during activation, consolidation periods, or during the short-term follow-up. Only minor complications were encountered, such as tooth injury (unerupted third molar), which was reported in one case (5%), and did not reveal any complications throughout the follow-up period.

Table 7
Analysis of mean changes for hard- and soft-tissue measurements (group 2).

	T2–T1			T3–T2			T4–T3			T4–T1		
	Mean	SD	*p*-value	Mean	SD	*p*-value	Mean	SD	*p*-value	Mean	SD	*p*-value
Skeletal measurements												
Horizontal												
B–VL (mm)	7.6	2.7	0.003	−0.2	0.4	0.4	−2.2	1.3	0.02	5.2	2.6	0.01
Pg–VL (mm)	10.6	2.4	0.0006	−0.6	0.9	0.2	−4.2	2.4	0.02	5.8	3.1	0.014
GO–Pg (mm)	13.6	2.5	0.0003	−1.4	1.3	0.08	−2.6	1.5	0.02	9.6	1.9	0.0004
Vertical												
ANS–Me (mm)	10.2	1.3	0.0005	−1.4	1.1	0.055	−2.2	1.9	0.06	6.6	1.3	0.0004
Ar–GO (mm)	18	1.2	0.00001	−2.4	1.7	0.03	−2.8	0.8	0.002	12.8	1	0.0005
MP–HL (°)	−11	1.6	0.0001	4.2	2.3	0.014	1.2	1	0.07	−5.6	1.7	0.002
Ar–GO–Pg (°)	−12.6	8.3	0.03	3.4	1.7	0.01	3	1.4	0.009	−6.2	5.8	0.07
Soft-tissue measurements												
Horizontal												
Bs–VL (mm)	11.8	2.4	0.0004	−3	0.7	0.0007	−2.4	2	0.06	6.4	1.5	0.0007
Pgs–VL (mm)	10.2	2.9	0.002	−0.4	0.5	0.2	−3.2	1.3	0.005	6.6	3.6	0.02
Li–E line (mm)	−4.4	1.1	0.001	0	0	1	1	1	0.09	−3.4	1.7	0.01
G–Sn–Pgs (°)	−14.2	1.3	0.0005	2.4	0.9	0.004	2.8	0.08	0.002	−9	1.7	0.0003

*Significant comparison, $p \leq 0.05$.

Table 9
Analysis of mean changes for hard- and soft-tissue measurements (group 3).

	T2–T1			T3–T2			T4–T3			T4–T1		
	Mean	SD	*p*-value	Mean	SD	*p*-value	Mean	SD	*p*-value	Mean	SD	*p*-value
Skeletal measurements												
Horizontal												
B–VL (mm)	12	5.6	0.0005	−0.62	1.2	0.2	−2.3	1	0.0005	9.1	5	0.001
Pg–VL (mm)	21.5	6	0.0005	−1.12	2.5	0.23	−4.8	4.4	0.02	15.6	6	0.0002
GO–Pg (mm)	12.06	5.3	0.0004	−0.31	0.7	0.24	−2.12	0.6	0.05	9.6	5	0.001
Vertical												
ANS–Me (mm)	9.3	4	0.0004	−0.62	1.2	0.2	−1.8	1.2	0.004	6.9	4.1	0.002
Ar–GO (mm)	11	6.8	0.003	−1.4	2	0.1	−2	0.9	0.0005	7.6	5.8	0.008
MP–HL (°)	−8.4	4.1	0.0007	1.3	1.8	0.08	2.12	1.2	0.002	−5	3.2	0.003
Ar–GO–Pg (°)	3.7	9.2	0.3	1	1.9	0.2	0.62	0.7	0.06	5.3	7.3	0.08
Soft-tissue measurements												
Horizontal												
Bs–VL (mm)	14.8	4.7	0.0005	−1.8	1.5	0.01	−2.4	0.9	0.0002	10.6	4.5	0.0003
Pgs–VL (mm)	20	6.9	0.0003	−1.4	1.3	0.02	−2.6	2.3	0.02	16	6.7	0.0003
Li–E line (mm)	−7.6	2.1	0.0005	0.4	0.7	0.2	1.6	0.7	0.0005	−5.6	2.6	0.0004
G–Sn–Pgs (°)	−18	6.5	0.0001	1	1	0.03	2.8	1.8	0.003	−14.3	5.5	0.0002

*Significant comparison, $p \leq 0.05$.

Table 10
Degree of patients' satisfaction.

	T2–T1	T3–T2	T4–T3
Esthetic	0	0	0
1 (Not satisfied)	0	0	0
2 (Slightly satisfied)	0	0	0
3 (Moderately satisfied)	0	0	0
4 (Satisfied)	0	3 (15%)	8 (40%)
5 (Completely satisfied)	20 (100%)	17 (85%)	12 (60%)
Function			
1 (Not satisfied)	0	0	2 (10%)
2 (Slightly satisfied)	0	0	0
3 (Moderately satisfied)	0	0	0
4 (Satisfied)	3 (15%)	5 (25%)	6 (30%)
5 (Completely satisfied)	17 (85%)	15 (75%)	12 (60%)

In the same patient (5%), during the preparation of a drilling hole for interosseous wire fixation of the genial segment, the drill bit fractured, and it was inaccessible to be removed. A decision was made to leave the drill bit in its place and follow up for any adverse reactions. A transient paresthesia of the lower lip was noted in all the cases (100%) which completely resolved by the follow-up at the end of the first year. All the unilateral cases (60%) felt transient pain in the contralateral joint during the first days of activation, which decreased with time. In the bilateral cases (40%), an AOB developed during the activation phase and was corrected by using elastics and manual training. Five patients (25%) had erythema and tenderness around the distractor pins, which was managed with antimicrobial ointment and systemic antibiotics. At the end of the follow-up, only one major complication occurred in two patients (10%), which related to recurrence of the ankylosis. Details of the complications are presented in Table 11.

Table 11
Summary of complications.

Complications	Number of patients (%)
Minor	
Tooth injury	1 (5%)
Fracture of drill bit	1 (5%)
Paresthesia	20 (100%)
Pain	12 (60%)
AOB	8 (40%)
Erythema and tenderness	5 (25%)
Major	
Re-ankylosis	2 (10%)

Moreover, the results revealed that three patients suffered from OSA (included in group 3). The AHI and the average oxygen saturation were 18, 20, and 16 and 85, 89, and 86, respectively. After the release of the ankylosis, none of the patients showed further difficulty in breathing. The snoring symptom disappeared after performing MDO. The postoperative average oxygen saturation was more than 95%, and the AHI was less than 5. There was also an increase in the width of the oropharyngeal airway that was documented by lateral cephalometry (Fig. 3). After the long-term follow-up period, no recurrence of OSA symptoms was observed.

4. Discussion

Recently, DO became an important reconstructive procedure for the management of maxillofacial deficiencies. One of the major advantages of DO is that the surrounding soft tissues can also be expanded, resulting in an adequate stability. Despite its widespread use, there is still a lack of information regarding the long-term stability of the hard and soft tissues of hypoplastic mandible secondary to TMJ ankylosis. It was reported that the posttraumatic TMJ ankylosis due to road traffic accidents and falls is more common in developing countries (Elgazzar et al., 2010; Zhu et al., 2013), than was reported in this study. This could be because these patients were from a less educated population without easy access to maxillofacial surgeons to treat the condylar injuries. Unilateral ankylosis has been reported to be more common than bilateral, with a ratio of 1.5:1 (Bello et al., 2012; Babu et al., 2013); the same ratio was observed in this study. The percentage of females (70%) was greater than that of males (30%). This female predominance is probably due to the greater interest and motivation for facial esthetics in females, as reported by many authors (Erbe et al., 2011; Paunonen et al., 2018). In accordance with Shaughnessy et al. (2006) and Erbe et al. (2011), the mean age of our patients was 20.5 ± 3.4 years. It was concluded that a positive facial appearance plays an important role in the professional and private life of patients aged 20–30 years. This does not conflict with the fact that those patients are not only looking for improvement in their facial esthetics but also looking for improvement in their jaw function, which is considered their prime concern, especially in developing countries.

Three methods of applying MDO were used for the treatment of mandibular hypoplasia secondary to TMJ ankylosis (Rao et al., 2004; Feiyun et al., 2010): simultaneous MDO with release of TMJ ankylosis, release of ankylosis followed by MDO, and MDO followed

by release of ankylosis. Simultaneous gap arthroplasty and MDO provides two main benefits, simultaneous improvement in mouth opening and correction of facial deformity (Dean and Alamillos, 1999; Papageorge and Apostolidis, 1999; Rao et al., 2004; Yu et al., 2009; Feiyun et al., 2010; Xu et al., 2015; Ma et al., 2019). However, it can result in an improper outcome of distraction owing to unpredictable vector direction, which leads to improper control of the mandibular segments during the distraction process, producing malocclusion. Furthermore, the proximal bone segment would improperly move in a backward direction, neutralizing part of the effect of DO. In addition, it is not easy to perform adequate physiotherapy during the distraction process, because of post-surgical pain, resulting in re-ankylosis or creation of a pseudo-joint at the distraction site (Lopez and Dogliotti, 2004; Kwon et al., 2006; Hegab, 2015).

On the other hand, patients with severe dentofacial deformities usually require a staged treatment protocol (Li et al., 2012; Zhu et al., 2013). Many authors usually start with MDO, followed by gap arthroplasty (Sadakah et al., 2006; Shang et al., 2012; Zhang et al., 2018). The reason for this selection is that the ankylosed TMJ acts as a fixed point that pushes the mandible in a forward direction. In addition, performing arthroplasty before distraction may carry the risk of re-ankylosis. However, in this study we preferred to start with the release of ankylosis followed by MDO, as recommended by many authors (Lopez and Dogliotti, 2004; Qudah et al., 2005; Kwon et al., 2006; Li et al., 2012; Mehrotra et al., 2012; Karamese et al., 2013; Zhu et al., 2013; Hegab, 2015), because the initial release of the ankylotic mass enables the patient to restore jaw movements, thus facilitating adequate nutrition, good oral hygiene maintenance, and preparation of the patient for the more complex MDO procedures. Moreover, during the first-stage procedure, the surgeon will be able to observe any malocclusion and recurrence of ankylosis that might influence the MDO treatment plan. However, the most important step in the successful treatment of TMJ ankylosis is the wide bone resection of the ankylotic mass, regardless of the sequence to be used when performing MDO and arthroplasty (Al-Moraissi et al., 2015).

The mean postoperative MIO in this study, following gap arthroplasty, was 40.2 ± 1.7 mm (range 30–45), which was maintained for 12 months before distraction planning. This is relatively consistent with the results of Papageorge and Apostolidis (1999) and Dean and Alamillos (1999). No evidence of re-ankylosis was observed in any patients during the short-term period. Unfortunately, two patients (without costochondral graft) developed re-ankylosis at the end of the follow-up. Similarly, Zhang et al. (2018) reported four cases (10%) of re-ankylosis within two years of initial treatment by MDO, followed by arthroplasty. The authors stated that the lack of physiotherapy was responsible for the recurrence, which was the main cause of re-ankylosis in our patients (10%). Therefore, it was recommended that the exercises should be continued for a minimum of six months and maintained for two years (Zhang et al., 2018; Khalifa, 2018).

Many authors support the stability of MDO results during the short-term follow-up (McTavish et al., 2000; Ko et al., 2004; Amano et al., 2009; Marsan et al., 2009; Baas et al., 2015; Vos et al., 2009) as reported in this study, although others revealed that relapse usually occurs in the short-term follow-up (Marquez et al., 2000; Rachmiel et al., 2001). In addition, Meazzini et al. (2005) and Shetye et al. (2006) reported that the majority of relapse occurs in the first year or three years post-distraction; after that, the results become stable. This is in contrast to our long-term results, which exhibited significant relapse at the end of the follow-up period; however, the facial improvement mostly remained.

MDO cannot completely improve the retruded chin that almost always exists in cases of TMJ ankylosis. In our study, 65% of the patients (groups 2 and 3) agreed to do an advancement genioplasty to improve their facial profile. In these cases, MDO failed to resolve the retruded chin completely, as reported by many authors (Li et al., 2012; Erbe et al., 2011; Khalifa and Mohamed, 2018). On the other hand, patients of group 1 were satisfied with their facial appearance following MDO and refused to undergo genioplasty. The mean chin advancement (Pg point) in this group was 5.6 ± 4.3 mm, which was produced by a clockwise rotation of the mandible during the distraction process (Khalifa and Mohamed, 2018; Koide et al., 2013; Venditelli et al., 2008). The advancement genioplasty (groups 2 and 3) resulted in a large horizontal advancement, which was observed at Pg (10.6 ± 2.4 mm; 21.5 ± 6 mm, respectively) when compared to advancing the mandible with MDO, as seen in group 1. In this study, B point was chosen because it is always above the genioplasty osteotomy line. It acts as an ideal reference point to differentiate between MDO advancement and advancement of the chin by genioplasty as reported by van der Linden et al. (2015). Patients who received genioplasty following MDO (groups 2 and 3) had a greater advancement in the horizontal direction (7.6 ± 2.7 mm, 12 ± 5.6 mm, respectively) at the B point versus 5.4 ± 2.6 mm in group 1. By the end of the follow-up, all groups showed significant relapse at the B and Pg points but did not reach the preoperative value.

In this study, all patients were classified as high-angle patients. A significant reduction in MP-HL° was reported in all groups (T2–T1), this was followed by a significant increase during the short-term period in group 2 only (4.2° ± 2.3°), then continued to increase significantly in all groups throughout the long-term follow-up. These results contradict the studies of Hamada et al. (2007) and El-Bialy et al. (2013) who reported that MP-HL° increased significantly during the distraction and remained almost constant during the long-term follow-up. However, Gonzalez et al. (2001) reported a 4.1° increase in MP-HL° during the distraction and 2.7° of further opening of this angle during the consolidation period. Further opening of MP-HL° was likely a result of muscular pull on the developing callus (Gonzalez et al., 2001) or due to poor compliance with the elastic chin-cup as was reported by El-Bialy et al. (2013). In our study, we used intermaxillary elastics at the end of the activation period to counteract the increase in the MP-HL° and control bite opening, as advocated by Wei et al. (2007). This vertical compression minimizes the suprahyoid muscles' pull and molds the immature regenerate, guiding the distal segment back into the proper occlusal position.

The effects of aging on soft-tissue changes have to be considered when analyzing the long-term effects of MDO on the lower lip and chin region. It was reported that from the age of 24–34 years, the nose grows forward, the lips retrude, and Pgs moves backward (Forsberg, 1979). This agrees with our findings when comparing the long-term data with that of the short-term. Joss et al. (2013b) also observed that an increase in the patient's age was significantly correlated with a downward movement of Pgs ($p < 0.05$) and to a backward movement of Li and Pgs ($p < 0.05$). In addition, Joss et al. (2013a,b) found a significant correlation between the amount of advancement at point B and the upward movement of Li. In our study; there was no significant correlation between the amount of relapse of all cephalometric parameters with the age and gender of the patients. These findings are in accordance with many authors (Baas et al., 2015; Joss et al., 2012). Joss et al. (2013a) found a significant correlation after MDO and at long-term follow-up for points Bs and B; this is in contrast to our findings in which an insignificant moderate correlation between points Bs and B at the long-term follow-up was found in all groups. On the other hand, an insignificantly moderate correlation was found between points Pgs and Pg at the long-term follow-up in group 1, whereas in groups 2 and 3, the correlation was found to be strong and statistically

significant. The Li point also advanced anteriorly toward the esthetic line, leading to an improved profile; this also was reported by many authors (Seifeldin et al., 2014; Khalifa and Mohamed, 2018).

The ratio between horizontal soft-tissue changes and skeletal changes was reported after MDO in many studies (Melugin et al., 2006; Joss et al., 2013a,b; Talebzadeh and Pogrel, 2001). In our study, the ratio between Pg: Pgs and B: Bs (T4–T1) in the three groups ranged from: 1:1.1 to 1:1.3. These ratios coincide with that reported by many authors (Asadi et al., 2014; Posnick et al., 2016; Khalifa and Mohamed, 2018). The previously reported ratios of soft- and hard-tissue movement vary greatly among studies because individual soft-tissue drape could vary and the behavior of the lower lip after correction may vary from one patient to another. Furthermore, genioplasty itself could change the vertical dimension of the hard and soft tissues (Ewing and Ross, 1992).

In the current study, the corpus length increased during MDO. The amount of lengthening was nearly equal to those in previously reported studies (Karacay et al., 2005; Batra et al., 2006; Aizenbud et al., 2010). There was an insignificant decrease in the length of the corpus during the short-term follow-up in all groups. However, it remained almost the same in group 1 and exhibited a significant relapse in groups 2 and 3 at the end of the follow-up. Minor skeletal relapse was also reported by many studies (Karacay et al., 2005; Sadakah et al., 2006; Aizenbud et al., 2010). A study by El-Bialy et al. (2013) reported that the increased corpus length remained stable throughout the long-term follow-up, as reported in group 1. The ramus height was increased significantly in the three groups after the distraction process. These results are coincident with those of Aizenbud et al. (2010). Throughout the long-term follow-up periods, the ramus height decreased significantly. This reduction in the effective bone length may account for the remodeling processes at the gonion and pogonion anatomical points because of the changes in the direction of the soft-tissue muscle pull on the mandible (Shetye et al., 2006; Aizenbud et al., 2010). Many authors found high rates of relapse in the corpus length (25%) and ramus height (26%–87%) from the length achieved by distraction (Marquez et al., 2000; Shetye et al., 2006). In the current study, we observed a much smaller relapse percentage at the end of the follow-up for the ramus height and corpus length when compared with those previous studies.

One limitation of the study is the treatment plan, which was based on clinical examination and lateral cephalometry, which is two-dimensional radiography, whereas the mandibular deformity secondary to TMJ ankylosis is multidimensional and requires three-dimensional virtual planning. The small number of patients who met the inclusion criteria could also be another limitation; there is a great need for long-term results in a larger sample size. Strengths of this study include the long-term follow-up of cases that had facial deformity due to TMJ ankylosis, which is a multidimensional deformity. In addition, it was a single-institution experience and evaluated multiple outcomes.

5. Conclusion

TMJ ankylosis leads to severe, multidirectional mandibular hypoplasia, which is significantly corrected with the MDO. The MDO provides a stable short-term improvement in the facial esthetics at the first postoperative year, but a significant relapse occurs during the long-term follow-up. Nevertheless, a satisfied facial esthetic is maintained for up to seven to 12 years postoperatively. During the activation period, the MDO minimizes the gained MIO after release of ankylosis, but the MIO is successfully restored with physiotherapy.

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Conflict of interest

No conflicts of interest.

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