A systematic review on the outcome of distraction osteogenesis in TMJ ankylosis

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ABSTRACT

Introduction: TMJ ankylosis (TMJa) management by arthroplasty alone fails to address the deformity and the compromised airway. Distraction osteogenesis (DO) can offer a pragmatic solution to TMJa. The aim of the study was to generate evidence towards the role of DO in TMJa, evaluate its efficiency and develop an algorithm for use of DO in TMJa.

Material and methods: The research question was formulated using the PICOS statement for reporting guidelines in systematic reviews, where the efficiency of DO was evaluated in terms of mouth opening, correction of facial deformity and asymmetry, airway correction, and its long term effects.

Results: 1130 articles reported DO as a treatment modality for TMJ ankylosis, of which 32 prospective studies, 16 retrospective and 2 RCTs were included in the study. DO was used for mandibular distraction in 45 studies and for simultaneous maxillamandibular distraction in only five studies. An algorithm for use of DO in TMJa was developed.

Conclusion: Although DO has proven its application in TMJ ankylosis cases, its best use is for correction of obstructive sleep apnoea. Relapse causing loss of posterior ramal height is a concern after transport DO. Prearthroplastic DO appears to best correct mandibular deformity. A maxillomandibular deformity requires simultaneous maxillomandibular distraction. However, a metaanalysis is still awaited for effectiveness of DO in TMJ ankylosis.

Temporomandibular joint (TMJ) is the only joint actively dealt by Oral and Maxillofacial surgeons and yet has been the center of controversy throughout these centuries. The spectrum of management of this joint in TMJ ankylosis (TMJa) ranges from gap arthroplasty to alloplastic total joint replacement. Even meta-analysis and systematic reviews have failed to project the best out of gap arthroplasty, interposition arthroplasty, autologous condyle reconstruction, distraction, or alloplastic replacements. Today, the goal is not just to achieve the function but also to restore the lost anatomy to the pre-disease level and allow normal growth. So, removing the whole condylar joint component and not reconstructing it may seem like a heinous crime in today's era.

Conventionally, the fused joint and micrognathia were managed by separate surgeries for ankylosis management and the standard reconstructive option was autogenous graft. Ideal requisites preferred for graft were its ability to assume a similar shape and form, should be sturdy enough to maintain it, has an adaptive potential to forces placed by

mastication and is dynamic to sustain the physiologic wear and tear for long term. The ideal reconstruction however should not only address the mandibular component and the diseased fossa but also restore complete form and function with prevention and addressal of the secondary facial deformity if any.

Distraction osteogenesis (DO) is a promising concept for this and has been used for reconstruction of new autologous bone that resembles condyle and regeneration of all attached soft tissue components creating along with a layer of intervening fibrous tissue that behaves as pseudo disc.¹ This regenerative technique of osteodistraction has found considerable application in correction of obstructive sleep apnoea and aesthetic correction of facial deformity by lengthening of bone and soft tissue and forming new bone of similar embryonic origin and character. The simultaneous lengthening of both hard and soft tissue provides physiologic environment and forces, that further facilitate remodelling and is thus considered a ray of hope towards achieving an ideal solution to this

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disease of joint fusion.^{1,2} This study aimed to generate evidence towards the role of DO in TMJa and develop an algorithm for use of DO in TMJa.

1. Material and Methods

The review question was to evaluate the outcome of DO in patients of TMJ ankylosis. The research question was formulated using the PICOS statement for reporting guidelines in systematic reviews, where the outcome of DO was evaluated in terms of mouth opening, correction of facial deformity and asymmetry, airway correction, and its long term effects. Randomized controlled trials (RCTs), clinical trials, and prospective and retrospective studies evaluating the clinical efficiency of DO in managing the TMJ ankylosis patients were included. Case series, literature reviews and expert opinions were excluded from the qualitative analysis in the present review.

Information sources and search: The standard electronic bibliographic databases 'MEDLINE (via PubMed)', 'EMBASE', 'Google Scholar', 'The Cochrane library', 'The Cochrane Central Register of Controlled Trials', 'LILACS', and 'Trip medical database' were searched from January 1, 1990 to December 1, 2020. The relevant data addressing the review question was searched using the following keywords and their combinations: "Distraction osteogenesis" (MeSH term) AND "Temporomandibular joint" (MeSH term); "Distraction osteogenesis" (MeSH term) AND "Ankylosis" (MeSH term); "Distraction osteogenesis" (MeSH term) AND "Temporomandibular joint" (MeSH term) AND Ankylosis (MeSH term); "Distraction osteogenesis" (MeSH term) AND "Temporomandibular joint" (MeSH term) AND "Arthroplasty" (MeSH term); "Distraction osteogenesis" (MeSH term) AND "Ankylosis" (MeSH term) AND Mouth opening (All terms). The reference lists of possible included studies were hand searched and articles addressing the review question were selected for full-text analysis. The search was extended to the trial registries and

grey literature as well to find the relevant articles. Experts were also contacted to obtain the relevant information through email to improve the robustness of the search strategy. The literature search was limited to the published articles in English only. The latest search for study selection was done on December 31, 2020. The study is submitted to PROSPERO (ID-259369) for registration.

Study collection and data extraction: Initial literature search (Titles and abstracts) was done by the two reviewers separately using the aforementioned key terms. Titles were assessed and the articles addressing the research question were separated for abstract analysis. Abstracts were further assessed and articles specifically addressing the review question were selected for full-text analysis. Data extraction from the included full-text articles was done and tabulated. **Studies with statistical data in terms of mean change of mouth opening, amount of distraction achieved or advancement achieved, asymmetry correction and relapse were segregated to be analysed quantitatively.**

2. Results

A total of 1130 articles were identified. The articles, in which DO was used as a treatment modality for TMJ ankylosis, were considered in the evaluation. After an extensive search, 50 clinical studies and 32 case series/case reports were retrieved. Case series were excluded due to non uniformity of evaluated parameters (Fig. 1) Clinical studies included 32 prospective studies, 16 retrospective studies and 2 RCTs (Fig. 2a). where DO found its application for mandibular distraction osteogenesis (MDO) in 45 studies and for simultaneous maxilla-mandibular distraction (SMMDO) in only five studies.

The two RCTs included were by Chellappa et al.,³ where prearthroplasty and simultaneous arthroplasty mandibular distraction was compared, and other by Kaur et al.⁴ who compared TDO with

Fig. 1. Prisma flow diagram.

Fig. 2a. Distribution of studies in different types of DO.

costochondral graft. A total of 729 patients were recruited from these 50 clinical studies. Mean change in mouth opening available in 13 of these studies was pooled in 276 patients. Out of these 13 studies, 11 had performed mandibular distraction. (3 each had prearthroplasty and post arthroplasty DO, 5 had simultaneous MDO, and 2 had TDO. Pooled mean change in mouth opening across 13 studies was found to be 29.16(95% CI - 26.84–31.46, $I^2 = 98\%$) with random effect model (Fig. 2b). Further stratification based on type of distraction could not be done due to lack of sufficient numerical data. As lesser number of studies were available for pooling of data for the outcome of mean change in the amount of distraction with advancement, asymmetry correction and relapse, these findings could not be analysed statistically. Earlier distraction was achieved using extra-oral distractors but with the advent of miniaturized distractors, the use of internal distractors was substantial amongst these studies (Fig. 2c).

2.1. Why distraction osteogenesis (DO) ?

With long-standing ankylosis especially in the growing phase, there is a deficiency of not only the bone but also of the soft tissues on that side. Though the face appears full on the affected side giving a false sense of the excess of tissues, the ipsilateral muscles are fibrosed. Hence an excision of the ankylotic mass may just not be sufficient even to restore the full range of mouth opening. Also, such a joint release may even shorten the posterior facial height.⁵ When a graft is used in such a situation to restore the ramal height, strong tension by the surrounding muscles may lead to relapse. So ideally beside achieving a stable mouth opening, a compensatory return of growth lag to correct deficient facial dimensions should also be considered.

DO can be beneficial in providing all the above requirements. An average post operative maximal mouth opening of 25–35 mm at follow

Fig. 2b. Pooled Mean change in mouth opening.

Fig. 2c. Intraoral Vs extraoral distractor.

Fig. 3. Types of DO in TMJ ankylosis.

up is considered as a successful outcome in TMJa.^{2,4} The pooled estimate of studies in this manuscript for which findings were available also reported the mean change in mouth opening to be 29.16(95%

CI - 26.84–31.46, I2 = 98%). This random effect pooled estimate confirms the satisfactory results of DO in general terms of outcome of mouth opening in cases of TMJa. Further, if this can be undertaken at a young

Table 1
Mandibular Transport Distraction studies in TMJa.

S. No	Author, Year	Type of Study	No of TMJa Patient	Age (mean) year	Type of Device Intraoral/Extraoral	3D Planning	Follow-Up (months)	Mean Change Mouth Opening (mm)	Mean Change Airway (mm)	Mean Advancement (mm)	Asymmetry Correction	Midline Deviation Correction	Complications
1.1	Cheung et al., 2007	Prospective	5	3–51	Extraoral	Yes, Splint	24 X ray	Preop 3–25 ¹⁴ Postop 38	NM	13	Mandibular deviation still present	NM	discomfort during activation ²
	Schwartz et al., 2008	Prospective	4	21–56	Extraoral	No	7–29	Improved ^{3–36}	–	13–30	Yes	Yes	None major
	Gabbay et al., 2006 [#]	Prospective	4	0–11 (6.8)	Extraoral	No	12.5	26.5 but 13.3 at 1yr	NM	28.5	NM	NM	48% relapse
	Eski et al., 2008	Prospective	3	14–17	extraoral	No	13, MRI	20–22	NM	5–10 antero-posterior, 11–17 vertical	Corrected	NM	Distractor failure ¹ MRI showed neocondyle
	Feiyun et al., 2010	Prospective	16	18–43	Intraoral 2 each side	Yes	39.7 X ray, CT, PSG	Preop 4.6, Postop 33.5	Improved airway	18.9 ramal length	Yes	NM	None
	Oncul et al., 2011	Prospective	8	17–33	Intraoral 4 Extraoral 4	No	24 X ray	Preop 1–8 Postop 30–41	NM	mean ramal length 39 Co-Go 13 Lat cephal, 18 PA cephal	Ratio of affected to unaffected ramus 0.95	Yes	NM
	Xiao et al., 2012	Retrospective	7	22.9	Intraoral	No	39.4 (16–92) CBCT	Preop 4.1 Postop 31.4	NM	Cond length decreased by 6 mm (10.5. to 4.5) later	Asymmetry increased 17% at follow up	NM	Condylar resorption. ⁴
	Sahoo et al., 2012	Retrospective	8	<1–42 (13.6)	Extraoral	No	12–24 X ray, CT	33.75 ± 1.58	NM	NM	NM	NM	Infection
	Bansal et al., 2013	Prospective	6	4–8	Extraoral	No	13–27, ¹⁹ CBCT	Preop 3.5 Postop 29.1	NM	12	NM	Yes	Pin loosening, ³ Transient marginal Mand N palsy, ¹ CBCT showed neocondyle
	Xu et al., 2015	Retrospective	18	20.4	Intraoral	No	37 (6–81) X ray, CT	Preop 7.1 Postop 32	NM	Mand length 12.3(6.8–21) Cond length 10.7 ^{6–17}	Yes	NM	Distractor break ¹
	Farina et al., 2017	Retrospective	12	5–30 (11.4)	Intraoral	No	36–156	33	NM	Bilateral Ramal ratio decreased from 1.27 to 1.07	Yes	6°	Reankylosis ² managed by gap arthroplasty
	Kohli et al., 2017	Prospective	11	18–48	Extraoral	No	6 CT	Preop 4.8 Postop 32	NM	Cond height width 9.9, 5/11 had resorption >7 mm	NM	NM	Transient facial nerve paresis, distractor site, hemorrhage, distractor device loosening

(continued on next page)

Table 1 (continued)

S. No	Author, Year	Type of Study	No of TMJa Patient	Age (mean) year	Type of Device Intra oral/Extra oral	3D Planning	Follow-Up (months)	Mean Change Mouth Opening (mm)	Mean Change Airway (mm)	Mean Change Advancement (mm)	Asymmetry Correction	Midline Deviation Correction	Complications
	Xia et al., 2020	Prospective	21 ²⁶	>14	Extraoral	Yes	>24, 34.1 ± 13.3	Preop 8.7 ± 5 Postop 31.2 ± 8.3	Improved airway	13.8 Ramus height 8.2 ± 4 Go-FHP decreased 6.2 ± 4	Yes	Yes	re-ankylosis ¹ , anterior open bite ⁵
	Kaur et al., 2020	RCT	12TDO	3–16	Extraoral	No	24–39, Ceph, MRI	Pre op 9.5 ± 7 Post op 34.4 ± 8.9	NM	10.42 distracted bone	Yes, asymmetry from 8.5 to 4, chin asymmetry correction 26% improved	Yes Midline deviation from 2.67–1.33	Non union, ¹ Infection pin site ¹ Need disc formed
	Ramly 2020	Retrospective	28(44)	0–14	Extraoral	No	14	21.4 ± 7.3	NM	NM	NM	NM	Recurrence rate 21/28 cases(75%) ⁵ tracheostomy ⁵ gastrostomy ⁵

age, it will surely counteract the associated psychological impacts. Above all DO is considered a safe method for the correction of facial deformities. Although minor complications do occur, they do not compromise the treatment outcome. Moreover, these complications can be prevented by proper virtual surgical planning and precautions.²

2.2. Types of DO in TMJa (Fig. 3)

2.2.1. Transport distraction osteogenesis (TDO) for condylar regeneration

Transport distraction osteogenesis is a movement of a bony segment of the ramus of mandible that is contoured superiorly to look like a condyle. It allows formation of physiologic bone biologically at the trailing edge without a bone graft and a fibrocartilaginous tissue at the leading edge. This fibrous tissue at the leading edge is prevented from contacting the opposing bone and thus performs the function of a pseudo-disc. This way TDO restores the posterior facial height and corrects mild facial asymmetry (Table 1).^{4,6–22}

Bansal et al.¹⁴ in a prospective study in 2013 reported no significant difference in cancellous bone density, trabecular number, or spacing in the neocondyle at 9 months postoperative CBCT. Eski⁹ confirmed the remodelling of leading-edge into neocondyle and the presence of mature fibrous tissue intervening in the space between the condyle and the glenoid fossa using MRI in 3 patients treated with simultaneous distraction. However long-term dimensional stability of neocondyles was questioned by Xiao et al.¹² based on 7 years post-operative evaluation on panoramic radiographs of seven unilateral ankylosis cases treated with TDO. They showed a statistically significant reduction in the achieved condylar height.

Shortcomings: TDO is a lengthy process in comparison to other reconstruction modalities like costochondral (CCG) or sternoclavicular grafts (SCG) which provide a single surgery solution. The risk of complications like pin tract infection and soft tissue or bone infection still exists. A small transport segment may at times undergo resorption. Also, it is technique sensitive procedure as vector control is challenging to ensure condylar reconstruction. Another limitation is active mouth opening exercises may get hampered which are mandatory after ankylosis release. High relapse rates of TDO up to 50% in TMJa is a drawback reported by various studies. Gabbay⁸ reported relapse of 48%. Xiao¹² reported change of achieved condylar length from 10.55 mm to 4.5 mm at follow up and change from 1.27 to 1.07 mm was reported by Farina.¹⁶

TDO vs CCG: Autologous graft especially CCG was considered the gold standard, especially in ankylosis management in growing children as it is biologically similar to the condyle and the cartilage cap provides the growth center. But it has been debated for its unpredictable growth, mismatch of medullary and cortical bone, inadequate strength of bone leading to fractures and deformation, the possibility of resorption or infection, all ultimately leading to compromised long term results or even recurrence besides their obvious donor site complications.

DO lengthened bone and soft tissue both abolished the need for graft and its associated morbidity. Experimental studies to compare distraction and CCG on the same animal showed that the neocondyle matched CCG and both the reconstructed condyles remodeled well with time and in long term behaved like condyles. A period of immobilization is required for the success of the grafts. The risk of failure of graft uptake exists due to the scarring which limits its use in recurrent cases where multiple surgeries are done. Unpredictability due to resorption/overgrowth of grafts or heterotopic bone formation is the biggest disadvantage.⁴ Most of the comparison studies have found distraction to be promising except a few.

TDO vs SCG: Kohli et al.¹⁷ compared 11 patients each of transport distraction and sternoclavicular graft for joint reconstruction, and found SCG to be superior in terms of better mouth opening. They found that greater condylar height and width could be achieved with SCG with lesser complications. Greater mean condylar resorption was noted in the DO group.¹⁷ According to Kohli et al. two major factors reported for excessive resorption of created neocondyle were distraction length

greater than the length of the transport segment and delay in mouth opening exercises that prohibits the required stimulus.

TDO vs TJR: Total prosthetic replacement (TJR) in TMJ ankylosis replaces all involved components like condyle, intervening tissue as disc and glenoid fossa. Customized TMJ-TJR has the advantage of accuracy, improved fit, and avoid any need to alter the bone to fit the prosthetic joint, and hence serves as the best alternative. However, complications like mechanical failure, wear debris leading to inflammatory reactions are known to happen. DO provides an autologous reconstruction without the side effects of the prosthetic joint.

As TJR does not grow, neither it allows growth, its use is controversial in the growing patients, even though it is currently being debated for use under 16 years of age. Correction of the occlusion can be achieved by permitting maxillary dentoalveolar growth to compensate the cant in both TDO as well as TJR. Early mobilisation is recommended in TJR as well as in staged DO, when ankylosis release is the second procedure. Also TJR in these patients requires virtual surgical planning to check the fit of the stock prosthesis, else a custom prosthesis may be required at the time of ankylosis release.

Gabbay et al.²⁶ compared transport distraction with Mathews device arthroplasty. Mouth opening in the DO group improved to a mean of 27.5 mm but in 12.5 months it reduced to 14.3 mm. In group I (transport distraction osteogenesis), the mean incisor opening was 1 mm preoperatively and 27.5 mm postoperatively; however, it relapsed to 14.3 mm by 12.5 months follow-up (48% relapse). Mean incisor opening in group II (Mathews Device arthroplasty) was 3.9 mm preoperatively and 33.4 mm postoperatively and remained at 30.6 mm after 11.1 months follow-up (8% relapse). One patient in group I (transport distraction osteogenesis) underwent surgical revision because of relapse. Data showed that for TMJ bony ankylosis both transport distraction osteogenesis and Mathews Device arthroplasty techniques were successful initially; however, the Mathews Device arthroplasty avoided long-term relapse.

2.2.2. Mandibular distraction osteogenesis for obstructive sleep apnoea (OSA) correction^{23–43,44–52}

Mandibular distraction is lengthening of mandible as a part of treatment strategy of TMJa. In patients of “Triad ankylosis” with a triad of obstructive sleep apnoea (OSA), reduced posterior airway space, and micrognathia, only ankylosis release will lead to further narrowing of airway because when a patient opens mouth small posterior placed mandible swings behind further pressing on to the airway, which can cause activation of trigemino-cardiac reflex. All this demotivates the child for physiotherapy precipitating chances for reankylosis.²⁷ MDO widens airway considerably and can minimize airway concerns.

Andrade³¹ advocated a protocol and proposed mandibular distraction first in cases with apnoea hypopnoea index (AHI) > 30 & posterior airway space (PAS) < 4 mm. Anchilia¹⁹ advocated mandibular distraction for AHI > 20. Preference for different protocols is differential among studies. Prearthroplasty distraction was preferred in severe deformity cases in which airway compromise and obstructive sleep apnea were prime complaints. Bi³⁵ assessed change in airway space on CT. A mean area increases of 54% and a volume expansion of 73% was seen. However, they have reported no significant difference between prearthroplasty and simultaneous distraction technique in terms of airway improvement after distraction in TMJ ankylosis cases. Hasan et al.⁴¹ on 20 patients of ankylosis with at least 7 years of follow-up also reported a significant increase of airway width similar to other studies.^{20,26,27,31,35,50}

2.2.3. Mandibular distraction osteogenesis for facial deformity correction

Arthroplasty for mouth opening is a mandatory need for TMJa patients, whereas aesthetic correction is a desirable need for poor patients. Hence, an aesthetic correction by prearthroplasty MDO before joint release provides a wholesome solution to the problem and with a better psychological boost. Also severe deformities yield better results with staged treatment.³⁷ The advancement achieved with DO restores the lost

posterior ramal height and corrects the retruded mandible, occlusal cant, and dental midline.

Advancement due to DO can be assessed at B point which is above the genioplasty osteotomy line. Studies have found that it improves facial esthetics considerably but according to them despite sufficient mandibular advancement, 65% of patients required genioplasty for ideal chin projection and deviation correction. Further improvement of advancement can be assessed for adequacy at Pg (Pogonion) point. This was also supported by Cheung 2007,⁶ Yu 2008,⁴⁴ Zhu 2011³⁷ etc.

Three accepted protocols for mandibular distraction have been followed in the literature with certain advantages and disadvantages.

Prearthroplastic DO^{3,23–35}: Pre-arthroplasty distraction is preferred for multiple reasons. It allows distraction against a stable fixed point, such that a true distraction of mandible occurs and that too in the desired direction only, and with a better vector control. After arthroplasty distraction vector is difficult to predict and control. In other protocols, some length from distraction is lost due to upward or backward movement of the condyle. Formation of the pseudo joint at the callus in simultaneous and post arthroplasty cases are reported which is obviously an advantage here.⁵³ (Table 2) However prearthroplasty DO has been found to be associated with more pain in the contralateral normal TMJ during the course of DO.³ Practically and ideally Prearthroplastic DO is the best answer to TMJa.

Post arthroplasty DO^{36–41}: Proponents of postarthroplastic DO believe that the chief complaint of the TMJa patients i.e. inability to open their mouths, should be addressed first. This judiciously allows for mouth opening physiotherapy, unlike simultaneous distraction in which aggressive mouth opening may disrupt the callus. After ankylosis release, the actual growth deficit and malocclusion can be assessed and corrected using DO. Also patient compliance for physiotherapy can be encouraged, and recurrence can be minimized if DO is undertaken later as a second surgery.²⁰ (Table 3) Post arthroplasty DO does not limit the amount of distraction, unlike prearthroplasty distraction where occlusion becomes the limiting factor. It is important to note that although prearthroplasty distraction increases the mandibular ramal height, the ankylosis release surgery later causes some shortening of the ramus again.

Simultaneous arthroplasty and distraction (SAD)^{3,35,42–52}: It was first performed in 1999 by Papageorge and Apostolidis.²¹ Simultaneous arthroplasty and distraction was preferred as it allowed mouth opening by fused joint resection, reduced airway resistance, reconstructed ramal height, and corrected asymmetry by lengthening of the mandible in a single surgery (Table 4). A comparison of Prearthroplasty and simultaneous distraction showed that although prearthroplasty DO provides better control of the distracting segment, but unlike simultaneous distraction, it bears a disadvantage that the final mandibular position cannot be predicted as the final mandibular position tends to change due to muscle pull after the release of ankylosis.

2.2.4. Simultaneous maxillomandibular distraction (SMMDO)^{26,30,34,37,41}

An occlusal cant which exceeds 3° in frontal plane is clinically significant. In cases where delay in treatment leads to significant maxillary hypoplasia, mandibular distraction can be coupled with simultaneous maxillary distraction by Lefort 1 osteotomy and fixation at the contralateral zygomaticomaxillary buttress. Piriform buttress should be used when the amount of distraction is more.³⁰ This simultaneous maxillomandibular distraction will simultaneously lengthen the maxilla on the ipsilateral side along with the distraction of the mandible. It corrects the upper lip positioning as well as the occlusal cant, preserving the pre-existing occlusion and avoiding any worsening of occlusion.

2.3. Why should DO be preferred ?

In severe airway compromise, distraction even before joint release can widen the airway to obviate the need for tracheostomy. This decrease in airway resistance is most appreciated in three-dimensional volume rather than 2D parameters. The anatomic region showing a maximum

Table 2
Prearthroplasty MDO studies in TMJa.

S. No	Author, Year	Type of Study	No of Patients (Joints)	Age (years)	Type of Device Intraoral/ Extraoral	3D Planning	Follow-Up (months) Outcome CT/MRI/X ray	Mean Change MO (mm)	Mean Change Airway width (mm)	Mean Advancement (mm)	Asymmetry Correction	Midline Deviation Correction	Complications
5.1	Cohen et al., 1998	Retrospective	1TMJa, rest others	6–18	Extraoral	No	NM None	NM	AHI change 7.4 to 1.3, RDI from 25.2 to 1.7	22	NM	NM	Aspiration, pin site infection, pin loosening, transient nerve palsy
	Wang et al., 2003	Prospective	21	21.2	Intraoral	No	18.1 X ray	NM	Preop 4.6 Postop 12.5 AHI from 58 to 3.2	9–30, height from 60 to 70.8	Yes	NM	None major
	Ko et al., 2004	Prospective	1	1	Extraoral	No	10–26 X ray	5.8	NM	8.3, height gain 5.8	Minor asymmetry	from 9.8 to 1.6	Relapse of 30% in ramus height
	Sadakah* et al., 2006	Prospective	9	14–35	Extraoral/ Intraoral	No	17 X ray	34.7 ± 2.0	Preop 4.1 Postop 11.2 ± 1.2	20.7 ± 2.6 Maxillary length from 48 to 57	Occlusal cant 0–1°	Yes	None major
	Anantanarayanan, 2008	Prospective	3 (bilat 2 unilat 1)	8–12	Extraoral	No	X ray	NM	Mean change in ODI 24.2	13.8	Yes	NM	NM
	Elgazzar 2012	Retrospective	11	Adult	Extraoral	No	14–96 X ray, CT	34.7	NM	NM	NM	NM	Major and minor (33%)
	Yadav et al., 2014	Prospective	15	2–46	Extraoral	Yes	X ray, CT	NM	Min O2 saturation 81.2–98.2%, AHI from 57.03 to 6.7	15(22)	NM	NM	Infection at pin site ⁴ difficult intubation ¹ so tracheostomy
	Mehrotra et al., 2016*	Prospective	10	15–35	Extraoral	No	12–24 X ray, CT scan.	35.6	–	Ramal height 28.9	Occlusal cant Corrected	Yes	Non-union ¹ Transient involvement of facial nerve.
	Andrade et al., 2018	Prospective	25	6 - 17, 18- 26	Intraoral	Yes	12–28	Pediatric 4.64 ± 2.3 to 34.4 ± 2.3 Adult: 5.18 ± 1.8 to 33.9 ± 2.0	Pediatric: 3.6 ± 1.3 to 9.0 ± 2.4 Adult: 5.2 ± 1.9 to 10.9 ± 2.5	Pediatric: 10.07 ± 2.093 Adult: 9.36 ± 1.804	NM	NM	Infection ³
	Zhang 2018	Prospective	40	9–53	Extraoral 2 distractors	No	6–85 X ray, CT	Pre op 3.8 Post op 33	Improved oropharyngeal space	NM	Some Required surgery	orthogn	Relapse ⁴ pin side Infection ⁴
11.1	Kumar 2018	Prospective	10	10 ^{13–40}	Extraoral	No	60 X ray Epworth sleep scale	Preop 9 Postop 34	Preop 15.3 ± 3.77 Postop 6.20 ± 1.4 increased airway, snoring cessation	15.8	Yes	NM	Numbness lower lip(9) Open bite ²⁷
	Jiang 2018	Retrospective	7	7–12	Extraoral	no	24.8	30.5	NM	NM	Improved	Chin deviation decreased. from 8.16 to 1.32	None

(continued on next page)

Table 2 (continued)

S. No	Author, Year	Type of Study	No of Patients (Joints)	Age (years)	Type of Device Intraoral/ Extraoral	3D Planning	Follow-Up (months) Outcome CT/MRI/X ray	Mean Change MO (mm)	Mean Change Airway width (mm)	Mean Advancement (mm)	Asymmetry Correction	Midline Deviation Correction	Complications
13.	Shehata et al., *2007	Retrospective	5	14–22	Extraoral bidirectional distractor	No	1-5, X ray	NM	NM	14 ¹¹⁻¹⁸ Height 16 ¹³⁻²²	all cases	Corrected In all patients	None major, Transient pain during activation, Gingivitis due to IMF, Mental nerve paresthesia, ³ hypertrophic scars ³
14.	Chellappa et al. # 2015	RCT	10	5–30	Extra oral	No	15	Preop 5 Postop 34.3	NM	NM	In all cases	NM	Pain > in PAD at 1 month, Contralateral TMJ pain more No
15.	Bi et al. # 2018	Retrospective	5	>18	Intraoral	yes	5–12, X ray, CT	NM	3–125% improved	NM	NM	NM	No

*- SMMDO done, #- Two types of arthroplasty- PAD/SAD.

in-cre-ase of air-way was

Table 3
Postarthroplasty MDO studies in TMJa.

S. No	Author, Year	Type of Study	No of Patients (Joints)	Age (years)	Type of Device Intraoral/ Extraoral	3D Planning	Follow-Up (months) Outcome (CT/ MRI/X ray)	Mean Change Mouth Opening (mm)	Mean Change Airway	Mean Advancement (mm)	Asymmetry Correction	Midline Deviation Correction	Complications (No of patients)
1	Singh 2009	Retrospective	1 TMJa	4–15	Extraoral	No	12–24 X ray	NM	NM	28	NM	NM	Distractor failed, ¹ cellulitis ¹
2	Zhu et* al, 2011	Retrospective	5 (SMMDO 3)	>18	Intraoral	No	12–32 (18.6) X ray	Preop 3.4 Postop 32.5	Improved	NM	Yes	NM	Hematoma ²
3	Li et al., 2012	Prospective	12	17–27	Intraoral	No	8-36 (15.7 ± 2.8) X ray	Preop 3.3 Postop 35.8	Improved	12.9 ± 2.4	Yes	NM	Transient numbness temporal, chin, tracheotomy, ⁷ ant open bite ¹
4	Gupta et al., 2013	Prospective	14	14–22	Extraoral	No	NM	Maintained	Improved	14.2	Yes	Yes	None
5	Qiao 2017	Prospective	14	21–31	Intraoral	Yes	38–58	35.67 ± 3.39	Improved	6.17	Yes	Yes	None
6	Hasan *2019	Retrospective	20 ²⁸ 12 unilateral 8 bilateral (SMMDO 5)	Non growing adults	Extraoral	No	8.5 ± 1.5 X ray	32	Improved	46.7 ± 10.7	Yes	NM	Tooth injury, drill bit fracture left in situ, transient paresthesia lower lip, erythema, tender around pins, Recurrence(2)

Table 4

Simultaneous arthroplasty MDO studies in TMJa.

S. No	Author, Year	Type of Study	No of TMJa Patient (Joint)	Age (year)	Type of Device Intraoral/ Extraoral	3D Planning	Follow-Up (months)	Mean Change Mouth Opening (mm)	Mean Change Airway (mm)	Mean Advancement (mm)	Asymmetry Correction	Midline Deviation Correction	Complications
	Cascone 2002	Prospective	4	9–18	Intraoral	No	12 X ray	35	NM	16.5	Yes	Yes	None
	Rao et al., 2004	Prospective	6	7–10	Extraoral	No	24–36 X ray	28	NM	51 to 61.8	NM	NM	Anterior open bite ² swelling Scar pin site, Injury to unerupted tooth ¹ Minor discharge from pin site, ³ pin infection ¹
	Yu et al., 2009	Prospective	11	5–17	Extraoral 2, Rest Intraoral	Yes	34 ± 13 CT, X ray	Pre op 3.4 ± 1.4 32.4 ± 2.6 39.4	NM	12.4. ^{7–15}	Yes	Yes 2–3 mm overdone	postop hematoma ¹ No reankylosis.
	Mehrotra et al., 2009	Prospective	30	6–12	Both	No			NM	27.16	Yes	Yes	Localized infection, Pin loosening none
	Zhang et al., 2012	Prospective	4	14–23 ¹⁸	Intraoral	No	44–62 CT, X ray	Preop 0 Postop 33.5	NM	22.5. ^{20–25}	Yes	Yes	
	Egemen et al., 2012	Prospective	5	16–36	Extraoral	No	11.2 ^{6–21}	28	NM	NM	NM	NM	temporary frontal nerve weakness, ² intraop bleeding from Int Max artery ¹
	Khan et al., 2015	Prospective	8	Adult (NM)	Intraoral	No		Preop 6.25 ± 3.24	NM	Preop 39.50 ± 4.4 Postop 46.25 ± 3.1	Yes	Yes	Hardware failure ³
	Giraddi et al., 2016	Prospective	9	14.8	Extraoral	No	36 X ray	Preop 4.9 ± 1.8 Postop 34.9 ± 1.9	NM	Body 7.6 Ramus 7.4	7.5 (change Co-Go)	2.78 (midline), chin acceptable	No relapse infection at distractor site, ¹ Tooth injury osteotomy site
	Zanaty et al., 2016	Prospective	30	8–17	Intraoral	No	6 X ray Cormack Lehane score	38.8	ImprovedAHI 39.8 (38.8–40.9)	23	NM	NM	No complications
	Zhang et al., 2017	Retrospective	16	11–36	Extraoral	Yes	10–24 X ray, CT	Preop 3.6 Postop 32.8	NM	Increased body 13.6 Ramus10.6	Additionorthogn Surgery	Yes	No re-ankylosis observed in any patient
	Iqbal et al., 2020	Prospective	63	9–24 (17.3)	Extraoral	No	6	Preop-3.3 Postop-36.8	NM	Body 14 Ramus 10	Yes	Yes	Failure distractor ¹
	Chellappa [#] 2015	RCT	20 (SAD10)	5–30	Extraoral	No	15	Preop 5 Postop 35.9	NM	NM	In all cases	NM	Reankylosis ¹
	Bi et al. [#] 2018	Retrospective	17 (SAD12)	>18	Intraoral	yes	5–12 X ray CT	NM	3–125% improved	NM	NM	NM	No

(#- Two types of arthroplasty used, *- SMMDO used, 2 distractors used).

noted at the level of oropharyngeal (soft palate plane, increased by around 70%) and glossopharyngeal plane (back of the tongue, around 72%).³⁵

DO when performed at an early age minimizes the asymmetry development by preventing compensatory growth of normal side maxilla and occlusal canting. DO provides large elongation of the mandible, with soft tissue lengthening that gives stability, minimal relapse, and no donor site morbidity of graft harvesting. DO leads to elongation of the whole volume of muscle, unlike other reconstructive options where strained muscles tend to resist any acute elongation.

In DO some normal growth based on functional matrix theory is instituted and the neocondyle may allow growth as any normal mandible. Dynamic loading of the mandible by nearly normalized muscles also can promote enlargement of functional matrix and bone remodelling leading to match up of the normal growth on the unaffected side.⁴²

It restores and maintains the posterior vertical height, corrects asymmetry of the lower third of the face, and creates sufficient vertical and horizontal gaps to attain adequate occlusion. It helps to address micrognathia and gives projection to the chin. It creates TMJ articulation with the inferior lamina like a normal disc attached to the condyle.⁴⁴ It can be performed even in growing children where orthognathic surgery is not an option.

Farina⁵³ developed a “Rule of 10” based treatment algorithm that advocates distraction osteogenesis for all >10 years with severe asymmetries where the difference in height of both mandibular rami is greater than 10%.

Psychological benefits of distraction are also supported in the literature. Author themselves have evaluated and reported that distraction osteogenesis considerably improves oral health and health-related QoL in patients with TMJ ankylosis with facial deformities. The use of an external or internal distractor did not make any difference in the QoL; however, bilateral distraction, multivector distraction, and maxillomandibular distraction resulted in better QoL outcomes.⁵⁴

For every surgical procedure, long term stability is a prime concern. Relapse in MDO has been reported to be minimal, because of the gradual lengthening of the surrounding soft-tissue matrix.^{55,25, 56} However, many authors reported significant relapse during the follow-up period.^{26,56, 57} These controversies between results were present because different methods of evaluation and different timings have been used during follow-up. Meazzini⁵⁸ and Shetye⁵⁹ reported that the majority of relapse occurs in the first year or three years post-distraction; after that, the results become stable. Few authors^{59,60} found high rates of relapse in the

corpus length (25%) and ramus height (26%–87%) from the length achieved by distraction. Ko²⁵ concluded that mandibular length remained unchanged while facial height showed minimal changes but 30% relapse in ramal height was reported after distraction. This reduction in the effective bone length may account for the remodelling processes at the gonion and pogonion anatomical points because of the changes in the direction of the soft-tissue muscle pull on the mandible.⁵⁹ In contrast to this many short-term follow up studies of up to 1 year and longer follow up ones up to 5–6 years⁶¹ reported continuous growth and no relapse based on serial radiographs.

Reconstruction options can be chosen based on algorithm presented. For any patient with symptoms of Obstructive sleep Apnea and AHI>20 irrespective of age mandibular distraction that widens airway by elongating mandibular body can be attempted first. It is pragmatic even for neonates and infants. It obviates the need and complications associated with tracheostomy. In mild asymmetry cases where ankylosis must have ensued after or near growth completion restoring posterior ramal height by transport distraction or ramal osteotomy will correct further shortening of ramus caused by gap arthroplasty.

In cases with severe asymmetry age based correction is warranted. Severe deformity cases of age less than 15 years may need mandibular distraction while above 15 years of age with associated maxillary hypoplasia may require simultaneous maxillomandibular distraction. However, if there is a high palatal plane it seems best to manage the deformity with counter-clockwise rotation (CCWR) of whole maxillomandibular complex or combination of distraction/orthognathic with total joint replacement, as a single surgery can correct the deformity instead of the long treatment time as required with SMMDO. An algorithm for use of DO in TMJa has been developed and illustrated in Fig. 4.

2.4. How to perform DO

3D surgical planning: Traditional methods involve planning based on cephalograms, OPG, and CT scans. With rapidly developing 3-dimensional technology, conventional treatment planning has been replaced by virtual surgical planning on software like Proplan or Mimics innovation Suite (Fig. 5). A pre-operative model planning using a 3 D model also facilitates a better decision in terms of use of a monoplanar, curvilinear or multiplanar, internal or external, distractor by using model surgery to check the final position with the distractor activated.

Constructing a guiding template using rapid prototyping, allows accurate positioning of distractor intraoperatively to achieve the preoperatively planned vector. The 3D printed templates transfer the plan to the

Fig. 4. Algorithm for use of DO in TMJ ankylosis.

Fig. 5. a-b Virtual surgical planning for univector linear distraction in TMJ ankylosis patient.

actual surgical table, increasing the accuracy of results, reducing complications, and decreasing operative time, allowing faster healing and thereby early return to work. Further, it may be used as an educational tool not only for young surgeons but also for a better understanding of the patient. Also, postoperative CT 3D superimposition onto the preoperative CT data and virtual plan allows for evaluation of any deviation from the virtual plan.

Best esthetics with maximum results can be attained by virtual surgical planning. Out of 50, only 10 studies used stereolithographic models or osteotomy guides as an aid. Chen¹⁸ evaluated the accuracy of Virtual surgical planning in 7 adult cases of bilateral ankylosis and found the difference between the virtual plan and actual results was less than 2 mm ($0.64 \pm 1.90 \pm 0.85$ mm).

Ideally, the distraction vectors must lie parallel to the occlusal plane to prevent open bite from being produced during elongation.^{23,41,62} This holds true especially for anteroposterior distraction after the growth has ceased. In the growing adolescents, lateral open bite is of limited concern because of the maxillary dentoalveolar compensation with ongoing growth.

An angle between the long axis of the distractors and the maxillary occlusal plane should range from 30 to 40°. ⁶³ When vertical lengthening is planned an angle between the long axis of the distractor and the maxillary occlusal plane can range from 45 to 90°. ⁶⁴ Oblique vector is chosen to produce both vertical and horizontal advancement. The bilateral distractors should not be convergent, to avoid any strain on the temporomandibular joints. ^{65–67} Osteotomy is done perpendicular to the planned vector in any case. However, if an open bite occurs, early removal of distractor device and use of elastics for intermaxillary fixation helps in the moulding of the callus.

Selection of distractor: External distractors when multivector can

allow multidirectional movements but hypertrophic scars, pin tract infection are its drawbacks. The infection rate with extraoral pins ranges from 5 to 30%. External distractors with pins are less sturdy than submerged distractor devices. Their distraction rate also does not follow true 1:1 between the activation amount and bone separation. ⁶⁸ Internal distractors are more aesthetic but they allow only univector movement.

Crucial Steps for TDO: A gap arthroplasty is done through a preauricular incision to excise the abnormal ankylotic chunk and reduce the condylar height. The interposition of temporalis myofascial flap or buccal fat pad or dermal fat is done. Vertical sub-sigmoid or an inverted L osteotomy of posterior ramal segment is done and its superior surface is contoured as condyle. Vertical Osteotomy is preferred in cases with prominent antegonial notch, while in others an inverted L osteotomy can be performed. The vertical component of osteotomy is made 8–10 mm anterior to the posterior border. The horizontal arm of L-osteotomy is made at least 1 cm above the angle of the mandible. ¹⁹ If possible all osteotomy cuts in ankylosis cases should be done with piezoelectric saws as this causes less soft tissue damage and less risk of recurrence. ⁴ The cranial component is contoured to stimulate the glenoid fossa. Earlier flat gap osteotomies were preferred, and remodelling gradually changed the shape. With virtual planning and use of intraoperative jigs, contouring of fossa can be performed with accuracy. ¹⁸

Distraction is started after a latency period of 3–5 days until vertical elongation and projection of mandible as planned is achieved. Maintain a gap between the proximal distracted stump and the fossa. Check radiographically or close a few threads if needed. This prevents re-ankylosis and eventually allows soft tissue to mature to form a disc. Elastics may be used for moulding of the callus and correction of open bite. Post distraction orthodontics should be started at the earliest to settle occlusion.

2.4.1. Pertinent points for successful distraction

Meticulous and interdisciplinary planning including orthodontists preferably on 3D printed models or on virtual simulation is desired. Release of pterygomasseteric sling permits simpler ramus lengthening and less risk of early relapse. This can be simply achieved through retromandibular or submandibular approaches.

Gradual stretching of the surrounding soft-tissue matrix, less periosteal stripping, and positioning of the osteotomy site distal to the pterygomasseteric sling with minimal heat generation have been considered beneficial to the stability of the technique.⁶⁹ Ideal distractor alignment with precise distraction based on occlusal plane, chin projection, and dental midline act as guide to the amount of distraction. An over distraction helps to compensate for relapse and inadequacy of growth at reconstructed condyle.

Sadakah²⁶ reported a smaller degree of resorption of the reconstructed condyles and advocated that size of transport segment obtained from transverse osteotomy at the mandibular angle is larger than that from the L-shape osteotomy as there will be more vascular soft-tissue attachment and the blood supply will be adequate. Secondly, the distracted vertical distance may affect the long-term results. If the distracted distance is longer than or equal to the vertical dimension of the transport segment, the transport segment cannot contact the residual native ramus after the activation phase. The transport segment will lose the mechanical supports and blood supply from the native ramus, which may cause condylar resorption.

A mandibular plane angle is an important determinant of long-term stability of distraction besides the amount of advancement. Angles greater than 39° are associated with greater chances of relapse. Hasan⁴¹ reported a significant increase in mandibular plane angle both at short and long term follow up despite a good reduction in mandibular plane angle achieved during distraction. Gonzalez and colleagues⁶⁹ reported that an increase in mandibular plane angle was noted during the distraction which further opened during the consolidation period due to muscular pull on the developing callus. These results contradict the studies of Hamada⁷⁰ and El-Bialy⁷¹ who reported mandibular plane angle achieved during the distraction remained almost constant during the long-term follow-up. Intermaxillary elastics may be needed to prevent open bite formation and excessive increase in mandibular plane angle. Use of elastics to counteract this vertical downward pull by supra-omohyoid group of muscles, moulds the callus better and maintains occlusion.

Active physiotherapy not only ensures adequate mouth opening but also helps in the remodelling of the condyle and pseudo disc. Physiotherapy should continue months beyond distractor removal also.⁵ To further augment this remodelling and prevent overloading, a flat plane occlusal splint is recommended for 6 months after distraction.

Cortical outline on the distracted callus is an indicator of distractor removal. Cone beam CT can give a true volumetric assessment but cephalograms (PA, lateral) and orthopantomogram are alternatives for regular evaluation of the results. Dental midline can be assessed for the stability of distraction. An adequate consolidation phase can minimize these relapses. Occlusal stability is another key determinant for the stability of callus and the mandibular lengthening results. Splint fabrication, early orthodontics for ideal condyle fossa relationship, and stable muscular forces should be the aim in peri-distraction phase. In severe malocclusion cases, the occlusion cannot be settled after distraction just by orthodontic aligning. In such cases, additional orthognathic or prosthodontic procedures are warranted.

2.5. Limitations with DO

Mouth opening during the distraction phase is invariably restricted though it improves gradually with reattachment of muscles onto the ramus when the distractor is removed. Deviation during opening is inevitable due to absent lateral pterygoid muscle attachment to the distracted neocondyle. DO fails to resolve the retruded chin

completely.^{38,41,69,71,72} The procedure of DO is less invasive but is more technique sensitive as incorrect vector planning is not forgiving and cannot be modulated. It's a long treatment with multiple hospital visits. Postoperative infection leading to callus disruption and resorption leading to relapse is also reported. Scarring, damage to developing tooth buds, roots, device fracture, or failure are known potential problems.

2.6. Future directions

Most of the studies in this review have a short follow up of 6 months to 2 years. Long-term effects of the stability of achieved results or relapse are not known. Whether the results could match up the normal side growth or if further surgeries for recreated asymmetry due to growth are warranted is still unknown. Literature also has a paucity of information on the functional effects of neocondyle and the disc on TMJ function both on the affected and non-affected side on long term, which may require MRI studies with longer follow up.

Even in animal studies, mostly DO has been attempted with already preserved disc. Cheung et al.^{72,73} created TMJ ankylosis on goat model, surgically released it to compare distraction and CCG, and reported a viable transport bone segment till the end of distraction, followed by an initial resorption of the condylar end due to remodelling to form a smooth cortical outline. Cartilaginous cells especially type II collagen have been reported in the post distraction fibrocartilaginous pseudo disc in animal studies, however a long term appraisal of the nature of tissue formed in human studies is needed.

3. Conclusion

Although DO has proven its application in TMJ ankylosis cases, its best use is for correction of obstructive sleep apnoea or mandibular lengthening. Relapse causing loss of posterior ramal height is a concern if considering transport DO for neocondyle regeneration. Prearthroplastic DO appears to be the best timing for correction of dentofacial deformity in the mandible; if properly planned for distraction vector using virtual surgical planning. A maxillomandibular deformity can be successfully managed with simultaneous maxillomandibular distraction. However, a metaanalysis of all outcomes will provide the best evidence for effectiveness of DO in TMJ ankylosis.

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Declaration of competing interest

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Appendix A. Supplementary data

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