

Simultaneous Gap Arthroplasty and Distraction Osteogenesis in the Management of Unilateral Bony TMJ Reankylosis – A Case Report

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Abstract

Temporomandibular Joint ankylosis is the adhesion of the condyle of the mandible to the base of the skull. It is the most common cause of hypoplasia of the mandible and eventual facial asymmetry. This case report describes the management of unilateral bony re-ankylosis of temporomandibular joint after the failure of a costochondral graft in an eight-year-old girl. The management of temporomandibular ankylosis presents a major challenge due to its increased reoccurrence rate. In the present case, simultaneous gap arthroplasty and distraction osteogenesis were performed to separate the ankylotic mass and lengthen the ramus-condyle unit. The one year follow-up of the patient showed adequate mouth opening. We recommend concomitant gap arthroplasty and distraction osteogenesis for effectively managing temporomandibular joint bony ankylosis.

Keywords: Distraction Osteogenesis, Gap Arthroplasty, Temporomandibular Ankylosis

1. Introduction

Temporomandibular ankylosis is defined as the fusion of the mandibular condyle with the base of the skull which may be bony or fibrous leading to limited mouth opening. Ankylosis comes from the Greek word for joint stiffness. It is also known as craniomandibular joint ankylosis^[1]. The American Academy of Orofacial Pain (AAOP) defines TMJ ankylosis as a “restriction of movements due to intracapsular fibrous adhesions, fibrous changes in the capsular ligaments (fibrous-ankylosis) and osseous mass formation resulting in the fusion of the articular components (osseous-ankylosis)”^[2]. The etiological factor leading to TMJ ankylosis includes - trauma (13-100%), local or systemic infection (10-49%), or systemic disease (10%)^[3].

Recently, distraction osteogenesis has been the method of choice for the management of TMJ ankylosis^[1].

However, there are controversies regarding the best timing for performing distraction. Some authors lean-to restore the function of the jaw and correct secondary facial deformation later i.e., distraction after gap arthroplasty^[4], whereas others prefer first distraction later followed by gap arthroplasty at the time of distractor removal^[5]. Here we present a case of unilateral TMJ re-ankylosis managed with concomitant gap arthroplasty and intra-oral distraction osteogenesis as one stage treatment.

2. Case Report

An 8-year-old girl reported with the chief complaint of limited mouth opening and facial asymmetry (Figure 1a). Her parents revealed a history of trauma at 1 year of age for which no medical assistance was sought at that time. Eventually, there was a loss of function and the

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patient was on a soft diet. She was treated for limited mouth opening at 6 years of age with surgery. Clinical examination revealed facial asymmetry with retrognathia. There was a shift in the dental midline and chin deviation to the right. The patient was unable to open her mouth. On palpation of the preauricular region, movement of the right condyle could not be elicited. The maximal incisal opening was restricted to 2mm. Radiographic examinations of orthopantomogram and CT scan showed a bony unilateral right TMJ ankylosis with the elongated coronoid process and shortened ramus (Figure 1b and Figure 2a, 2b, 2c). A screw used in the previous surgery for attaching the costochondral graft was visible in the OPG.

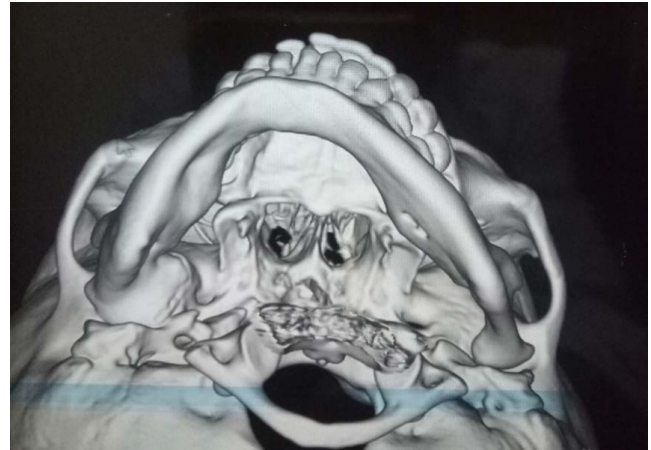


a

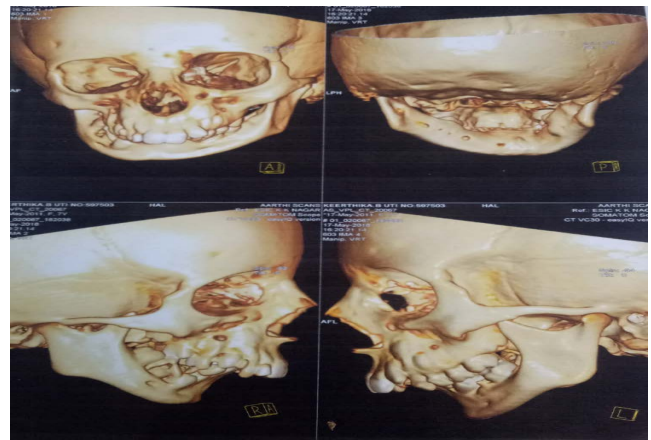


b

Figure 1. (a) Facial photograph preoperative, (b) Panoramic radiograph showed ankylosis of right TMJ with facial asymmetry and a screw placed for attachment of the costochondral graft in the previous surgery.



a



b

c

Figure 2. (a) Axial CT image, (b&c) 3D reconstruction shows severe bony fusion around the right TMJ with a deviation of the chin to the right side.

The protocol of Kaban, *et al.*, was followed in surgery. Gap arthroplasty and simultaneous distraction osteogenesis with bilateral coronoidectomy were performed. Gap arthroplasty was performed at the right TMJ through a preauricular approach. The aggressive resection of the ankylotic mass about 15mm was done with round bur and chisel. Glenoid fossa was recontoured and an ipsilateral coronoidectomy was performed. The maximum incisal opening obtained after the release of ankylotic mass and ipsilateral coronoidectomy was 24mm (Figure 3a). Contralateral coronoidectomy was carried out to increase the mouth opening. The maximum incisal opening obtained finally was 42mm (Figure 3b).

Distraction osteogenesis was performed with osteotomy cut in the angle via a retromandibular incision. Distraction was achieved by placing an intraoral

bone-borne unidirectional distractor (Figure 3c). When osteotomy was performed at the angle between 0° (the vertical vector) and 90° (the horizontal vector) with the occlusal plane, i.e., at the mandibular angle will result in an oblique vector. This will further result in vertical and horizontal elongation of the mandible. Extensive physiotherapy was carried for restoring normal TMJ function. The distractor was activated at a rate of 1mm per day, postoperatively after 5 days, and continued till midline was corrected (Figure 4). The distractor had been left in place for 12 weeks for integration. It was removed in the second surgery under general anesthesia. The patient was under follow-up for more than 1 year (Figure 5).

3. Discussion

Ankylosis of the TMJ is classified based on the site (intra-articular or extra-articular), the type of tissue affected (bony, fibrous or fibro-osseous tissue), and the degree of fusion (complete or incomplete)^[1]. It can be either true or pseudo-ankylosis. True ankylosis is a condition within the articular capsule, where there is osseous or fibrous adhesion between the surfaces of the TMJ. Pseudo-ankylosis is caused by diseases that are not related to the joint^[6].

The main etiological factors involved are trauma, infection, inflammation, and systemic diseases, with the

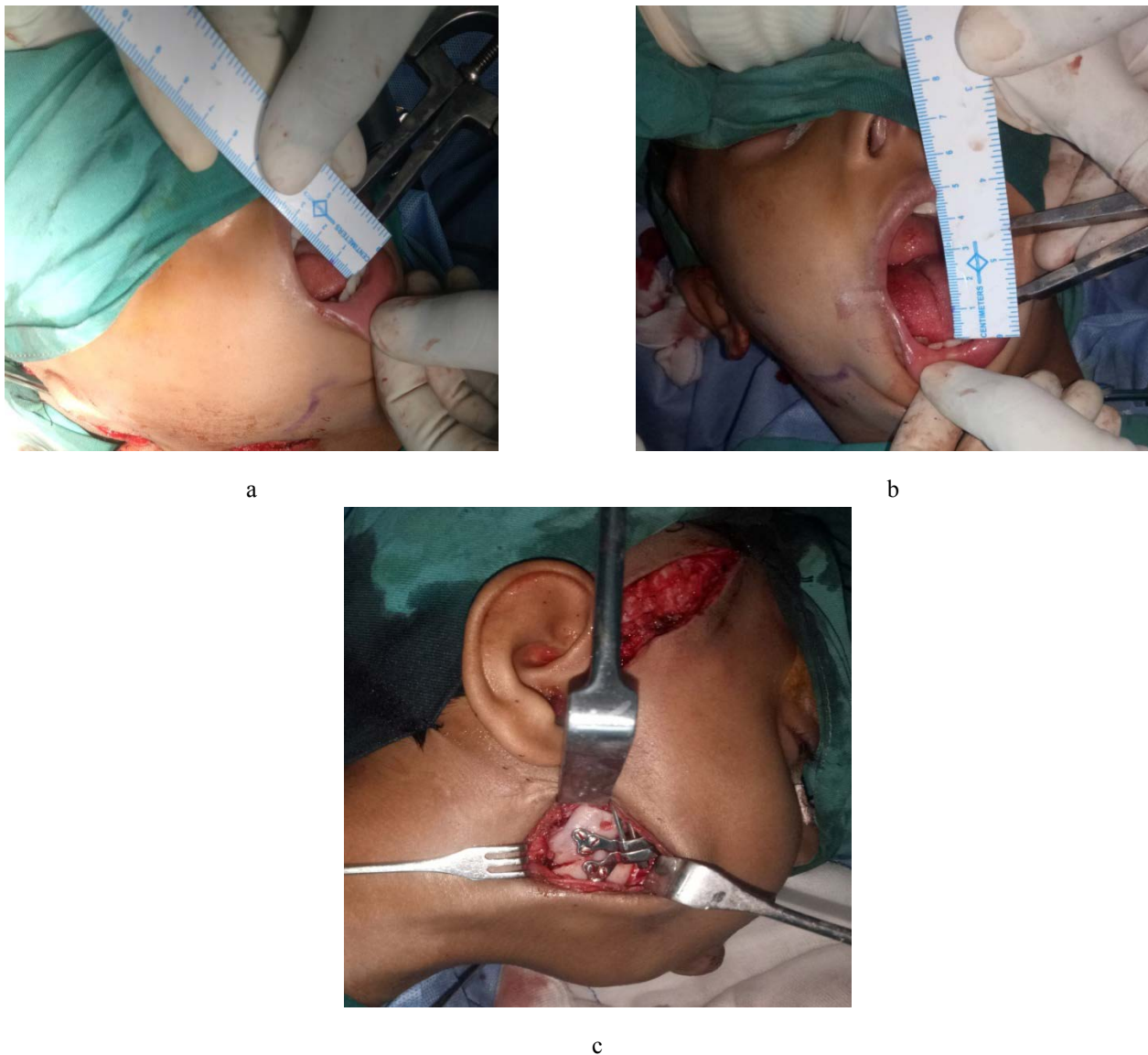


Figure 3. (a) Mouth opening after the release of ankylotic mass and ipsilateral coronoidectomy – 24mm, (b) Mouth opening after contralateral coronoidectomy- 42mm, (c) Distractor placed at the angle of the mandible through a retromandibular incision.



Figure 4. Panoramic radiograph after lengthening of mandible and correction of midline after distraction osteogenesis.



Figure 5. 1 year follow-up revealed mouth opening of 38mm.

first two being the leading causes^[7]. In the case of trauma, it is observed that intra-articular hematoma, along with scarring and formation of excessive bone, contributes to hypomobility. TMJ infection usually follows a contiguous spread from local infections such as otitis media and mastoiditis. It may also be caused by the hematogenous spread of infectious conditions or systemic infections such as tuberculosis, gonorrhea, or scarlet fever. The known systemic diseases to cause TMJ ankylosis includes ankylosing spondylitis, rheumatoid arthritis, psoriasis, sickle cell anemia, and fibrodysplasia ossificans progressive^[4,8]. TMJ surgery may also cause ankylosis. In

this case, the initial cause of ankylosis is due to trauma present at the age of 1 year.

In TMJ ankylosis, restricted movement of the mandible in children often leads to physical and social disabilities, including impairment of speech, chewing difficulties, dental caries, poor oral hygiene, facial abnormality^[9]. Furthermore, there can be airway obstruction with obstructive sleep apnea due to glossoptosis and reduced oropharyngeal space^[10]. Therefore, early diagnosis and management are crucial in reestablishing joint function and harmonious jaw function^[9]. This patient-reported with trismus/restricted mouth opening, mandibular deficiency with retrognathia, and lateral deviation of the chin to the right side.

The main objectives post-surgically are: a) to establish adequate/acceptable opening of the mouth, b) to completely excise the ankylotic mass for prevention of re-ankylosis in the long term, and c) to establish balanced facial esthetics to facilitate psychological development of the child. Different surgical techniques have been advocated for managing ankylosis with a facial deformity in children, which includes the release of ankylosis with gap arthroplasty, inter-positional arthroplasty, genioplasty, orthognathic surgery, and distraction osteogenesis^[11].

In 1990, the 7 step protocol was proposed by Kaban, *et al.*^[12] for systematic management of TMJ ankylosis. It includes – “(1) aggressive resection of the ankylotic segment, (2) ipsilateral coronoidectomy, (3) contralateral coronoidectomy when necessary, (4) lining of the joint with temporalis fascia or cartilage, (5) reconstruction of the ramus with a costochondral graft CCG, (6) rigid fixation of the graft, and (7) early mobilization and aggressive physiotherapy”. In 2009, this protocol was further modified, in which distraction osteogenesis was suggested as an alternative modality to costochondral graft for condylar reconstruction^[13].

TMJ reconstruction following ankylosis release is a challenging task due to its unique anatomy and physiology. Autogenous costochondral grafts are widely used for its growth potential and its biologic compatibility. Though costochondral grafts are the gold standard modality for the reconstruction of TMJ in children, it involves second surgical site exploration with possible complications of surgical site infection, chances of graft failure, and unpredictable growth of the graft^[1,9]. Various alloplastic materials are available to overcome these drawbacks. The above-reported case is of recurrent complete, bony unilateral true ankylosis with failure of the costochondral

graft. In this case, the patient was treated at 6 years of age with gap arthroplasty and costochondral graft to correct the facial deformity. The failure of the costochondral graft and re-ankylosis lead to restricted growth of the mandible and mouth opening. Besides the restricted growth of mandible, the patient suffered from severe deformities in function and aesthetics that adversely affected her development socially and psychologically.

Traditionally, orthognathic surgeries have been considered as the preferred technique for the management of facial asymmetry. Vertical sliding ramus osteotomies have been used to reconstruct the ramus-condyle unit. The main disadvantage is the difficulty of extremely stretching the soft tissues^[11,14]. Further, interpositional material and a bone graft may be needed to prevent re-ankylosis and to fill the bony defect created after the superior repositioning of the segment.

Recently, Distraction Osteogenesis (DO) has emerged as a more comprehensive treatment modality to overcome the limitation of orthognathic surgeries such as the amount of facial asymmetry which can be corrected and the deficiency of soft tissue. Distraction osteogenesis is a gradual process in which new bone is formed in the surgically constructed gap. The controlled force of low magnitude applied results in gradual stretching of the surrounding soft tissue stimulates tissue growth^[15]. This reduces the chance of re-ankylosis. The major advantage over autogenous graft is it obviates the need for graft, elimination of the second surgical site^[1]. The main disadvantage is the long duration of the treatment and the need for the cooperation of patients and their families in activating the device.

In 1999, Papageorge and Apostolidis were the first to perform gap arthroplasty and mandibular DO simultaneously for the management of ankylosis micrognathia, which improved the opening of the mouth and mandibular lengthening^[16]. When this procedure was performed simultaneously, it reduced treatment time and additional surgery required. Yoon and Kim^[17] presented two successfully treated cases: unilateral ankylosis of TMJ with gap arthroplasty and distraction osteogenesis with intraoral mandibular distractor after the failure of the gap arthroplasty and interpositional costochondral graft. Rao et al suggested that distraction osteogenesis may be used for the correction of deformities of the mandible, concurrently with gap arthroplasty in patients with unilateral TMJ ankylosis^[18]. Conversely, after the release

of ankylotic mass, the disadvantage of this procedure is that the changes in the position of the mandible cannot be fully controlled in the distraction period. This can lead to re-ankylosis if there is no appropriate physical therapy^[10]. In this patient, concurrently gap arthroplasty and distraction osteogenesis with bilateral coronoidectomy were performed, following Kaban's protocol to improve TMJ function and correct facial deformity.

In 1995, McCarthy, *et al.*, was the first to introduce Intraoral Devices for mandibular distraction^[19]. Yoon, *et al.*, used intra-oral distractor through an extra-oral approach for simultaneous distraction and gap arthroplasty^[17]. Xu, *et al.*, suggested the use of a single preauricular incision with a penetrating distraction port at the preauricular region, for gap arthroplasty and distraction^[20]. In the present case, ankylosis was released with preauricular incision and through a retromandibular approach, the distractor was placed intraorally for lengthening the ramus-condyle unit.

Compared to orthognathic surgery, DO increases both ramus height and length of the body of the mandible and the functional movements can be started immediately after surgery as there is no need for intermaxillary fixation^[9]. In this case, the osteotomy cut was placed at the angle of the mandible, such that DO increases both ramus height and length of the body of the mandible.

It has been hypothesized that reactive elongation of the coronoid process happens as a sequel to hyperactive temporalis muscle owing to an insufficient translation of condyle in ankylosis^[21]. In the present case, both coronoid processes were elongated superiorly. A bilateral coronoidectomy was performed for improving mouth opening to 42mm.

After surgery, a latency period of 5-7 days allows time for initial callus formation and healing of the soft tissues. The consolidation and quality of bone formation are determined by the rate and rhythm of the distraction^[9]. In the present case, a gradual distraction rate of 1mm per day was done. It is necessary to undergo intensive physiotherapy to further strengthen the opening of the mouth, mostly to prevent re-ankylosis. The patient was of adequate age to comply with post-operative physiotherapy care. On more than 1 year follow-up, the patient's mouth opening 38mm was maintained. In this case, the simple exercise by the use of a wooden spatula was undertaken.

4. Conclusion

Though mouth opening improved with various treatment modalities, facial deformity and re-ankylosis remained a problem. Distraction osteogenesis serves as a superior modality of treatment for the repair of facial deformities. In conclusion, the management of bony ankylosis of TMJ with facial asymmetry and retrognathia can be effectively managed by simultaneous gap arthroplasty and distraction as a single-stage treatment procedure. Effective post-operative care at an early stage, intense physiotherapy, and rigorous long term followup are essential to prevent reoccurrence of TMJ ankylosis.

5. References

- Chugh A, Pasricha N, Bedi RS, Pandey P. Management of temporomandibular joint ankylosis in children: Current perspective. *Asian Journal of Oral Health and Allied Sciences*. Apr-Jun 2011; 1(2).
- Cunha CO, Pinto LM, de Mendonça LM, Saldanha AD, Conti AC, Conti PC. Bilateral asymptomatic fibrous-ankylosis of the temporomandibular joint associated with rheumatoid arthritis: A case report. *Braz. Dent. J.* 2012; 23:779-782. <https://doi.org/10.1590/S0103-64402012000600025>. PMID: 23338276.
- Leonard B, Kaban, Carl Bouchard, Maria J. Troulis: A protocol for management of temporomandibular joint ankylosis in children. *J. Oral and Maxillofacial Surg.* 2009; 67:1966-1978. <https://doi.org/10.1016/j.joms.2009.03.071>. PMID: 19686936.
- Babu S, Parmar, Utsav U, Bhatt, Shilpi U, Bhatt. Distraction osteogenesis following gap arthroplasty to correct facial asymmetry - A defined protocol in treatment of patients with temporomandibular joint ankylosis. *International J. of Healthcare and Biomedical Research*. October 2014; 03(01):117-124. www.ijhbr.com. ISSN: 2319-7072.
- Zhang W, Yang X, Zhang Y, Zhao T, Jia J, Chang S, et al. The sequential treatment of temporomandibular joint ankylosis with secondary deformities by distraction osteogenesis and arthroplasty or TMJ reconstruction. *Int. J. Oral Maxillofac. Surg.* 2018; 47(8):1052-1059. <https://doi.org/10.1016/j.ijom.2018.01.022>. PMID: 29706239.
- Kazanjian VH. Temporomandibular ankylosis with mandibular retrusion. *Am. J. Surg.* 1955; 90(6): 905-10. [https://doi.org/10.1016/0002-9610\(55\)90721-2](https://doi.org/10.1016/0002-9610(55)90721-2).
- Hameed H, Alamgeer, Shad S, Din Q. Etiology, clinical and radiographic features of temporomandibular joint ankylosis. *Pak. Oral Dental Journal*. 2013; 33:26-30.
- Fonseca RJ. *Oral and maxillofacial surgery: Temporomandibular disorders*. Philadelphia (PA): W.B. Saunders Company; 2000. p. 309-313.
- Sharma R, Manikandhan R, Sneha P, Parameswaran A, Kumar JN, Sailer HF. Neocondyle distraction osteogenesis in the management of temporomandibular joint ankylosis: Report of five cases with review of literature. *Indian J. Dent. Res.* 2017; 28:269-274. https://doi.org/10.4103/ijdr.IJDR_292_13. PMID: 28721990.
- Xingna YU, Jianing Wang, Shida Hou, Rongsheng Zeng. Mandibular distraction osteogenesis in the treatment of pediatric temporomandibular joint ankylosis with micrognathia and obstructive sleep apnea syndrome: A case report with 4 year follow up. *Experimental and Therapeutic Medicine* 2019; 18:4888-4892. <https://doi.org/10.3892/etm.2019.8119>.
- Giraddi GB, Arora K, Sai Anusha AJ. Distraction osteogenesis in the treatment of temporomandibular joint ankylosis with mandibular micrognathia. *Ann. Maxillofac. Surg.* 2016 Jan-Jun; 6(1):68-74. <https://doi.org/10.4103/2231-0746.186125>. PMID: 27563611, PMCID: PMC4979347.
- Kaban LB, Perrot DH, Fisher K. A protocol for management of temporomandibular joint ankylosis. *J. Oral Maxillofac. Surg.* 1990; 48:1145. [https://doi.org/10.1016/0278-2391\(90\)90529-B](https://doi.org/10.1016/0278-2391(90)90529-B).
- Kaban LB, Bouchard C, Troulis MJ (2009). A protocol for management of temporomandibular joint ankylosis in children. *J. Oral Maxillofac. Surg.* Sep. 67(9):1966-1978. <https://doi.org/10.1016/j.joms.2009.03.071>. PMID: 19686936.
- Su-Gwan Kim, Sun-Sik Park. Incidence of complications and problems related to orthognathic surgery. *J. Oral and Maxillofac. Surg.* 2007; 65:2438-2444. <https://doi.org/10.1016/j.joms.2007.05.030>. PMID: 18022466.
- Natu SS, Ali I, Alam S, Giri KY, Agarwal A, Kulkarni VA. The biology of distraction osteogenesis for correction of mandibular and craniomaxillofacial defects: A review. *Dent. Res. J. (Isfahan)*. 2014 Jan; 11(1):16-26.
- Papageorge MB, Apostolidis C. Simultaneous mandibular distraction and arthroplasty in a patient with temporomandibular joint ankylosis and mandibular hypoplasia. *J. Oral Maxillofac. Surg.* 1999; 57:328-333. [https://doi.org/10.1016/S0278-2391\(99\)90683-3](https://doi.org/10.1016/S0278-2391(99)90683-3).
- Yoon HJ, Kim HG. Intraoral mandibular distraction osteogenesis in facial asymmetry patients with unilateral temporomandibular joint bony ankylosis. *Int. J. Oral Maxillofac. Surg.* 2002; 31:544-548. <https://doi.org/10.1054/ijom.2002.0297>. PMID:12418572.
- Rao K, Kumar S, Kumar V, et al. The role of simultaneous gap arthroplasty and distraction osteogenesis in the management of temporomandibular joint ankylosis with mandibular

- deformity in children. *J. Cranio. Maxillofac. Surg.* 2004; 32:28-42. <https://doi.org/10.1016/j.jcms.2003.07.003>. PMID: 14729049.
19. McCarthy JG, Schreiber J, Karp N. Lengthening the human mandible by gradual distraction. *Plastreconstrsurg.* 1992; 89:1-8. <https://doi.org/10.1097/00006534-199201000-00001>.
20. Xu J, Long X, Cheng AH, Cai H, Deng M, Meng Q. Modified condylar distraction osteogenesis via single preauricular incision for treatment of temporomandibular joint ankylosis. *J. Craniofac. Surg.* 2015; 26:509-511. <https://doi.org/10.1186/s40902-016-0058-0>. PMID: 27014663, PMCID: PMC4778145.
21. Srivastava H, Hegde K, Nair P, Gharote H, Gupta N, Chand PH. Temporomandibular joint ankylosis - A case of double trouble. *International Journal of Contemporary Medicine Surgery and Radiology.* 2016; 1(1):21-24.
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How to cite this article: Vanmathi VK, Parasuraman V and Vanmathi R. Simultaneous Gap Arthroplasty and Distraction Osteogenesis in the Management of Unilateral Bony TMJ Reankylosis – A Case Report. *Int. J. Med. Dent. Sci.* 2021; 10(1): 1945-1951.