ORIGINAL ARTICLE

Management of Obstructive Sleep Apnea and Non Apneic Snoring with Maxillo-Mandibular Distraction Osteogenesis

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Abstract

Background: Recently Distraction Osteogenesis (DO) has been used successfully for advancing maxilla and mandible to treat sleep disordered breathing like Obstructive sleep apnea and non apneic snoring. Prime advantage of DO is slow stretching of soft tissues and bone formation allowing greater advancement and enhancement of airway volume.

Patients and Methods: 10 cases with sleep disordered breathing due to maxillo-mandibular discrepancy of varying age groups were treated by various DO techniques.. All the cases showed gross improvement not only in facial and dental aesthetics but also respiratory symptoms. A mean increase of 5.14 mm and 1.85mm with respect to posterior airway space and retro palatal space was observed in mandibular corpus lengthening cases. All the cases showed improvement subjectively and objectively.

Conclusion: DO has been found to be valuable and gives us the ability to both prevent and correct the development of sleep disordered breathing.

Introduction

Sleep disordered breathing affects all age groups and the most common of them are non apneic snoring and obstructive sleep apnea. It is the most common yet often under appreciated conditions in Cranio-facial deformities. Maxillo-Mandibular distraction osteogenesis is one of the most contemporary and effective surgical treatment options [1] [2] [3].

OSA is a potential life threatening disorder caused by partial or complete obstruction of upper airway. If the obstruction sites are retroglossal or retro palatal maxillo-mandibular advancement would prove beneficial. Certain craniofacial abnormalities are susceptible to OSA and non apneic snoring. These include hypoplastic mandible, sagittal maxillary deficiency, narrow maxillary arch, long face syndrome, inferiorly and posteriorly positioned hyoid bone. Distraction Osteogenesis (DO) can be performed in all age groups and is a valuable tool that gives us the ability to both prevent and correct the development of sleep disordered breathing [1] [2] [3].

Distraction Osteogenesis is a biologic process of new bone formation between vascularised margins of bone segments gradually separated by incremental traction [4]. The basic concept of the procedure is induction of new bone formation along the vector of pull obviating the need for a bone graft. Finally, the increase in resulting bone stock will provide more reconstructive options in the future for traditional orthognathic surgical techniques.The traction force generates tension in the callus that connects the bone segments and this in turn stimulates bone formation. Distraction force also creates tension in the soft tissues including blood vessels, ligaments, cartilage, muscles, nerves which initiate a

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sequence of changes termed as distraction histeogenesis. Prime advantage of DO is slow stretching of soft tissues and bone formation allowing greater advancement than standard techniques. Advancing the maxilla and mandible in this manner will allow the enhancement of the upper airway volume gradually. This is similar to the titration accomplished when using adjustable mandibular advancement devices.

A study was carried out at our centre to assess the effectiveness of DO in OSA and non apneic snoring. The objectives were to compare results objectively and subjectively before and after the procedure.

Patients and Methods

All patients with severe maxillo-mandibular discrepancy with OSA or non apneic snoring undergoing DO for bilateral corpus lengthening, mid face advancement, transverse maxillary expansion during the last 39 months were considered as prospective study candidates (n=10). Clinical examination, PA and lateral cephalometric studies were performed according to established protocol for all the patients. Polysomnography (PSG) was done in cases with severe mandibular hypoplasia and respiratory symptoms. Three dimensional CT scan was also done in cases operated for TMJ ankylosis and syndromic cases. Comprehensive treatment plan based on the data base addressing facial aesthetics, occlusion and respiratory symptoms were formulated.

The treatment protocol followed in 8 cases included orthodontic preparation, distraction surgery, orthodontic therapy during distraction and consolidation, and post consolidation orthodontic treatment for settling the occlusion. In two young children with Treacher-Collins syndrome, DO was carried out without orthodontic intervention. The distraction surgery in all the cases was performed under GA.

The facts taken into consideration for building up the data base, both pre and post therapy, included age, sex, diagnosis, type of distraction and distractor device used, degree of lengthening, improvement on Apnea Hypopnea Index (AHI), Pre and Post treatment Posterior airway space (PAS) - Retro palatal space (RPS) are summarized in table 1.

Table 1 : Patient data, Procedure details and Results

Case NO	AGE/ SEX	DIAGNOSIS	Type of distraction osteogenesis	Type of distractor used	Clinical out come	Max/mand sagittal advancement	Posterior airway space pre/post(mm)	Retro palatal space pre/post (mm)	AHI(pre/ Post)/Resp Symptoms
1.	15 F	Optd bilateral TMJ ankylosis, hypoplastic mandible, severe OSA	Corpus lengthening	IO medicon body distracter	Good improvement in profile, chin projection. Satisfactory final occlusion	46/60	6/12	5/7	58.6/14.4 Cessation of snoring
2.	23 F	Optd bilateral TMJ ankylosis, hypoplastic mandible, severe OSA	Corpus lengthening	IO medicon body distracter	Satisfactory improvement in profile, chin projection with anterior open bite	52/70	8/11	6/8	48.2/20.6 Cessation of snoring
3.	22 M	Optd bilateral TMJ ankylosis, hypoplastic mandible, severe OSA	Corpus lengthening	IO medicon body distracter	Excellent improvement in profile, chin projection and occlusion	49/70	4/11	6/8	69.8/5.4 Cessation of snoring
4.	03 M	Treacher collin syndrome. Tracheostomy dependent	Corpus lengthening	Zurich paediatric distracter	Excellent improvement in profile, chin projection	33/48	3/10	4/6	Decannulated
5.	18 M	Class 2 Div 2 skeletal malocclusion with intractable snoring	Corpus lengthening	IO medicon body distracter	Excellent improvement in profile, chin projection	67/74	9/11	7/8	Cessation of snoring
6.	14 F	Maxillary hypoplasia Secondary to cleft palate surgery with severe snoring	Mid face advancement	Rigid external distracter	Excellent improvement in profile.Good final occlusion	82/98	11/11	1/7	Cessation of snoring
7.	22 M	Optd bilateral TMJ ankylosis, hypoplastic mandible with severe OSA, CPAP dependent	Mandibular Corpus lengthening	IO medicon body distracter	Excellent improvement in profile.Good final occlusion	55/69	5/12	6/8	Weaned of CPAP
8.	18 M	Severe transverse maxillo mandibular deficiency with snoring	Surgically assisted rapid maxillary expansion	Toothborne Hyrex appliance	Excellent improvement in facial aesthetics, occlusion	22/34 : inter molar width change	11/11	8/8	Cessation of snoring and mouth breathing
9.	16 f	Skeletal class II div 1 Malocclusion (mandibular deficiency) snoring oral breather	Corpus lengthening	IO medicon body distracter	Excellent improvement in profile chin projection, occlusion	72/79	8/12	7/8	Cessation of snoring and mouth breathing
10	6 M	Treacher collins syndrome	Corpus lengthening	Zurich paediatric distracter	Excellent improvement in profile chin projection, occlusion	41/57	5/9	5/7	Excellent improvement objectively &subjectively

Pre and post cephalometric data with respect to PAS, RPS, and mandibular length was compiled as per established protocol and was considered for this study [5].

Results

The patient data and results are summarized in Table 1. All the cases in our series showed improvement in facial aesthetics, occlusion and respiratory symptoms. A mean increase of 5.14mm and 1.85mm with respect to PAS and RPS respectively was observed in cases with bilateral corpus lengthening. In one case of mid face advancement the RPS increased by 5mm and snoring completely ceased. In the case with transverse maxillary deficiency, surgically assisted rapid maxillary expansion resulted in 12mm increase in inter molar distance, severe crowding could be relieved and patient reported complete cessation of snoring. One case of severe OSA due to severe mandibular hypoplasia and was CPAP dependent was weaned of CPAP following bilateral corpus lengthening with DO [6]. A three year old child tracheostomy dependent since birth was decannulated one month following DO (Fig 1,2,3).

Discussion

One of the most important indications for maxillo-mandibular distraction osteogenesis is severe OSA as a result of mandibular hypoplasia or maxillary deficiency. This is because DO addresses the main obstructive sites

Fig 1: Pre treatment frontal photograph of a three year old male child with Treacher –Collin syndrome and tracheostomy dependent since birth.

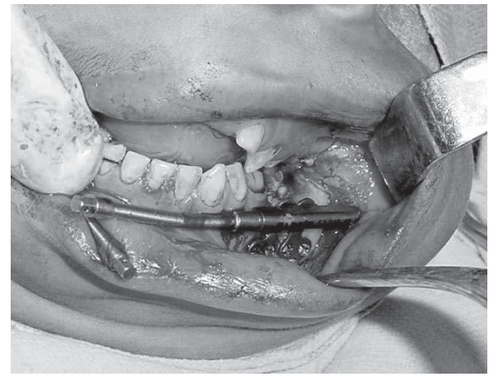

Fig 2: Bilateral mandibular distraction with Zurich Pediatric distracter.

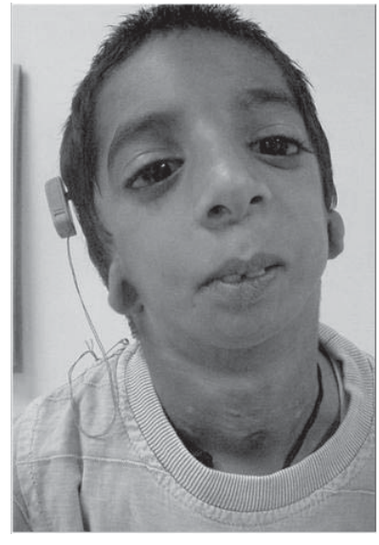

Fig 3: Post treatment photograph of the case following decannulation.

i.e, PAS and RPS increase in the 3-dimensions. Rachmiel and his co-workers analyzed bilateral mandibular distraction with compromised airway by 3D CT scan and concluded that DO of hypoplastic mandible, volume of hypoplastic mandible and upper airway increases eliminating the symptoms of OSA and preventing tracheostomy [7]. Although traditional orthognathic surgery are effective in treating mild and moderate OSA, the results are disappointing in severe OSA and when BMI >28 and in syndromic cases. This is because a traditional osteotomy has a physiological limit of approximately 10 mm of advancement which may be insufficient to open the upper airway for effective relief of obstruction in the obese patient or in those with severe disease. In the present series, we achieved a mean

advancement of 14 mm and in one of the severe OSA case we achieved 21 mm (Figs 4,5,6) .This is indeed the reason for significant improvement in respiratory symptoms. We have reported statistically highly significant ($p<0.001$) increase in total anterior face height and corpus length and excellent soft tissue response to large mandibular advancement with intra oral bilateral mandibular distraction osteogenesis [8] [9]. The cases considered in this study also consisted of cases of mandibular deficiency without respiratory symptoms [8].However we are of the view that all mandibular deficiency cases are potential OSA and advancing the mandible vide DO would not only improve masticatory function and facial aesthetics but also prevent OSA in the future.

In our series we followed the same protocol as recommended in the literature [10] [11] [12]. But we often used functional appliances like mandibular advancement device prior to and after corpus lengthening [8]. In cases of non apneic snoring associated with mandibular deficiency, use of mandibular advancement appliance during pre distraction orthodontic stage for neuromuscular adaptation showed reduced or ceased snoring. This was an indication to the treating team that surgically advancing the mandible would eliminate snoring. Pre-distraction orthodontics involved placing the dentition ideal to the skeletal bases using fixed orthodontic appliances and adjuncts. The highlights of the procedure involves performing osteotomy and placement of distracters (Figs 7,8).In case of mandibular corpus lengthening, osteotomy cuts were placed distal to the existing last molar. Lefort I level osteotomy was done for maxillary advancement and the same surgery coupled with mid palatal split was done for surgically assisted rapid maxillary expansion (SARME). Latency period of 5 days was followed; rate of DO was 1mm/day and rhythm of DO was 0.5mm/12 hr/day. Inter arch elastics were used during DO to mould the regenerate [13]. Respiratory symptoms gradually improved as the distraction progressed and all the cases showed gross improvement in the respiratory symptoms following completion of distraction. In a recently published article, Baur and Helman are of the opinion that the amount of DO can be individualized for each patient by performing PSG after 15-18 mm of advancement. If the PSG demonstrates a normal AHI, DO can be stopped to allow for consolidation. If AHI is elevated DO continues for 25mm [14].

Fig 4: Pre treatment Lateral Cephalogram of 21 year old male with severe mandibular hypoplasia secondary to growth disturbance following TMJ ankylosis and severe OSA. Note the gross reduction in PAS.

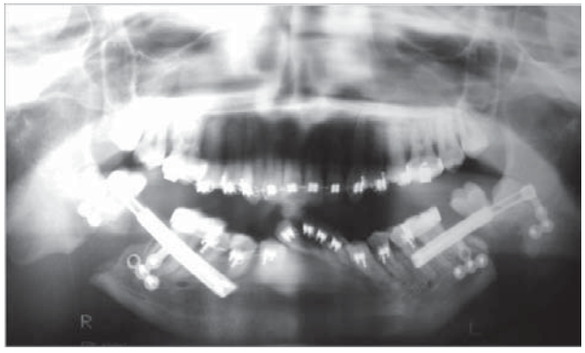

Fig 5: Ortho pantomo gram of the same patient at the end of distraction.

Fig 6: Post treatment lateral cephalogram showing gross improvement in facial profile , mandibular size and PAS.

Fig 7: Intra oral photograph showing Osteotomy distal to last molar and intra oral body distracter.

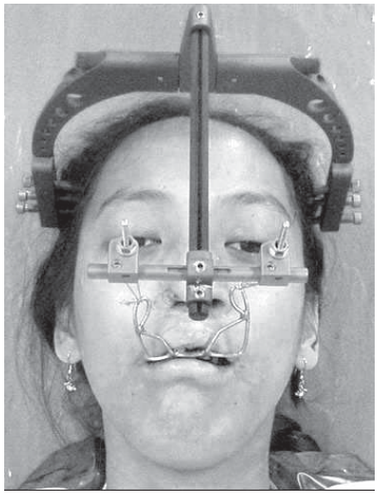

Fig 8: Rigid external distracter on a patient undergoing maxillary advancement.

Fig 9: Pre treatment lateral cephalogram of a 14 year old with maxillary hypoplasia resulting in concave facial profile and grossly inadequate RPS.

Maxillo-mandibular DO anteriorly positions the maxillary and mandibular frame work and their attending muscular attachments. In addition to addressing the obstructive sites namely, retroglossal and retro palatal regions, provides additional tension for genioglossus muscle and increases the available room in the floor of the mouth for the tongue and also improves tongue posture. Literature recommends maxillo-mandibular advancement as the most effective treatment of OSA other than tracheostomy and the success rate approach 100% [15] [16]. This procedure has been reported to have high degree of effectiveness in patients who are not successful with first phase surgery and who undergo this procedure even though they do not have maxillo-mandibular deficiency [17].

Studies with mandibular advancement devices has shown that there is increase in the size of pharyngeal airway at both post palatal and post lingual airway dimensions although greater increase is at the former [18].We have observed the same in corpus lengthening cases.

Oral appliances have been found to be the most acceptable for non apneic snoring, mild to moderate OSA [19] [20]. But in the present series non apneic snoring was associated with severe maxillo-mandibular discrepancy and the patients had sought consultation primarily for the later .In addition to gross improvement in facial profile and dental aesthetics , snoring completely ceased in these cases (Table1) [8] [9] .

Breathing difficulties during sleep may occur or exacerbate following repair particularly pharyngoplasty and palate repair in cleft palate children [21].In the present series, one case with severe maxillary deficiency secondary to operated cleft lip-palate and associated snoring was treated comprehensively with orthodontics, alveolar grafting and mid face advancement using rigid external distracter. Her RPS was found grossly inadequate (1mm) which increased to 6 mm following mid face distraction and snoring completely ceased and there was no deterioration in speech either (Figs 9 &10).Recently, the Pillar procedure has been successfully used for snoring and OSA. In this procedure polyester inserts were implanted into patient's soft palate to cause stiffening effect and thereby allow air to flow more freely through the patient's airway [22]. This procedure is likely to gain more acceptances in non cleft cases in the future.

Cistulli P has reported that OSA patients who have narrow maxillae and under gone palatal expansion either

with orthodontic appliance or by surgery show significant Improvement in the number of respiratory events that occur following expansion [23]. We have observed the same with complete cessation of snoring in one of our case who under went surgically assisted rapid maxillary expansion for severe constriction of maxilla and intractable snoring (Fig 11,12).In this case the PAS and RPS were with in normal limits. There is a reduction in nasal resistance and expansion at anterior nares following rapid maxillary expansion, therefore improvement in breathing [24]. A recent study to compare the effects of rapid maxillary expansion and surgically assisted rapid maxillary expansion on nasal volume using acoustic rhinometric methods concluded significant increase in nasal volume and decrease nasal resistance in two groups [25].

The application of a continuous stretching force on the two bone segments through the use of a device

Fig 10: Post treatment lateral cephalogram showing gross improvement in profile and increase in RPS.

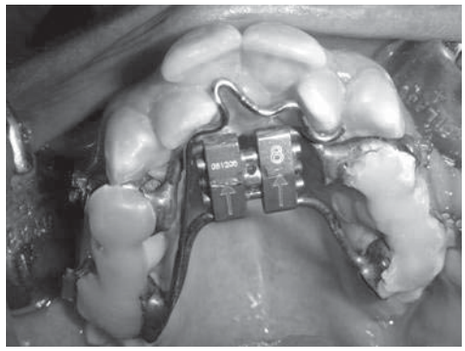

Fig 11: Pre distraction intra oral photograph of 19 year old male with transverse maxillary deficiency.

Fig 12: Post distraction intraoral photograph showing gross increase in maxilla in transverse plane.

triggers the conditions for growth. The undifferentiated cells in the bone marrow evolve into osteoblasts and begin formation of interlaced bone tissue orientated into parallel lines. Muscle and soft tissue mass increase via a process referred to as distraction histiogenesis. Clinically, this offers a distinct advantage as several craniofacial anomalies have soft tissue hypoplasia, in addition to deficient bony structures. This is especially effective in the stretch of the supra hyoid group of musculature along with histeogenesis in and around the tonsillar fossa with resultant effect in the palatoglossus and palatopharyngeous musculature resulting in an increase in the posterior pharyngeal airway space facilitating smooth and unlaboured breathing activity. Neurovascular elements contained within distracted bony segments also are stimulated to regenerate. In all our cases we found healthy new bone in the distraction zone both radiographically and clinically during the removal of the distractors. Osteocyte viability is essential to provide an adequate source of osteoblastic activity at the distraction site, hence careful surgical technique to minimize thermal or mechanical bone injury must be ensured. Similarly, an adequate blood supply to the distraction site & an intact periosteum and endosteum are critical to osteogenesis. In our surgical protocol, the bone cuts were restricted to osteotomy aimed at preserving the endosteal tissues thus facilitating an optimal bone healing [26].

OSA is common in a number of abnormalities associated with various congenital syndromes that involve mandibular hypoplasia(e.g. Pierre –Robin, Treacher Collin and Goldenhars syndrome) [27][28].Steinbacher and coworkers recently studied mandibular advancement by DO for tracheostomy dependent children with severe micrognathia. The results of this preliminary study indicate that mandibular advancement by DO is a

potentially viable treatment option for tracheostomy dependent children with upper airway obstruction secondary to micrognathia][29].We accomplished the same in one case (Table 1)

Distraction due to its versatility has established itself as a procedure of choice in a multitude of specific conditions. Cases treated with this technique show better stability with good quality of bone with long-term stability of the desired results.

Conclusion

DO for treatment of non apneic snoring and OSA caused by mandibular or maxillary deficiency is dependent on many factors including age of the patient. Unlike orthognathic surgery, DO can be applied in all age groups. The results were found to be stable and the respiratory symptoms in all the cases improved. It addresses the obstruction sites namely the retroglossal and retro palatal regions comprehensively and increases upper airway dimensions providing a 100% cure. The technique is indeed valuable and gives us the ability to both prevent and correct the development of sleep disordered breathing.

References

1. **Cohen SR**, Ross DA, Burstein FD et al. Skeletal expansion combined with soft tissue reduction in the treatment of Obstructive sleep apnea in children; physiological results. *Oto laryngol Head Neck Surg*. 1998. 119; 476-480.
2. **Diner PA**, Tomat C, Sergent B, Martinez H, Vanquez MP. Mandibular lengthening using intra oral distracters. In Craniofacial distraction Osteogenesis. Samchukov ML, Cope JB, Cherkasi AM. eds. Elsievier.2000; 247-255.
3. **Smith K**, Harmish M. Pediatric sleep apnea treated with distraction Osteogenesis. In Cranio facial distraction Osteogenesis. Samchukov ML, Cope JB, Cherkasi AM. eds. Elsievier.2000; 213-224.
4. **Cope JB**, Samchukov ML, Cherkasin AM. Mandibular distraction Osteogenesis: A historical perspective and future directions. *Am J Orthod Dento facial Orthop*. 1999. 115; 448-460.
5. **Borowiecki DB**, Kukwa A, Blanks RH. Cephalometric analysis for diagnosis and treatment of Obstructive sleep apnea. *Laryngoscope*.1998.98; 226-231.
6. **Menon S**, Roy Chowdhury SK, Vasant MR: Distraction osteogenesis in management of obstructive sleep apnoea. *Indian Journal of Oral & Maxillofacial Surgery,* 2003 .18 (3); 25-29.
7. **Rachmiel A**, Aizenbund D, Pillar G, Srouji S, Peled M. Bilateral mandibular distraction for patients with compromised airway analysed by 3D CT Scan. *International journalof Oral and Maxillofacial surgery*.2005.34(1); 9-18.
8. **Jayan B**, Roy Chowdhury SK, Kharbanda OP, Kotwal A. Orthodontics in intra oral bilateral sagittal mandibular distraction Osteogenesis. *Journal of Dentistry Defence Section*.2007. 2(2); 4-13.
9. **Jayan B**, Roy Chowdhury SK, Karkun S, Kamat UR.Treatment of skeletal class2 div2 malcclusion with associated upper airway sleepdisorder. A Case report. *Journal of Dentistry Defence Section*. 2006. 1(1); 46-49.
10. **Mazinni C**. Report on two international congress on distraction Osteogenesis. Paris and Stuttgart. 2005.
11. Distraction Osteogenesis . A new frontier in correcting Dentofacial deformities. Orthodontic dialogue of AAO. *Spring* 2000. 12(2); 1-4.
12. **Hanson PR**, Melugin MB. Orthodontic management of patients undergoing mandibular distraction Osteogenesis. *Semin Orthod*. 1999. 5(1). 25-29.
13. **Peltomaiki T**, Grayson BH, Venditelli BL, Katzen T, Mc Carthy JG.Moulding of regenerate to control open bite during distraction Osteogenesis. *The European Journal of Orthodontics*. 2002. 24(6); 639-645.
14. **Drew SJ.** Distraction Osteogenesis. sleep review; Journal for sleep specialists. May-Jun 2002.(www.sleepreviewmag.com).
15. **Prinsell JR**. Maxillomandibular advancement surgery in site specific treatment approach for obstructive sleep apnea in 50 consecutive patients. *Chest*. 1999.116; 1519-1529.
16. **Waite PD**, Shashidhar MS. Maxillo-mandibular advancement surgery : A cure for obstructive sleep apnea syndrome. *Oral and Maxillofac surg clinicNorth Am*. 1995. 7 ; 327-344.
17. **Li KK**, Riley RW, Powel NB, et al. Maxillomandibular advancement for persistent obstructive sleep apnea after phase I surgery in patients without maxillomandibular deficiency. *Laryngoscope*. 2000. 110; 1684-1688.
18. **Johal A**, Battagel JM. Current principles in the management of obstructive sleep apnea with mandibular advancement appliances. *British Dental Journal*. 2001.190(10) ; 532-536.
19. American sleep disorders association standards of practice committee, practice parameters for treatment of snoring and obstructive sleep apnea with oral appliances. *Sleep*. 1995. 18; 511-513.
20. **Jayan B**, Prasad BNBM, Rajput AK, Bhattacharya R, Dhiman RK. Management of obstructive sleep apnea with oral appliance: our experience. *Indian J Sleep Med*. 2006. 1(4). 204-207.
21. **Abramson DL**, Marrinan EM, Mulliken JB. Robin sequence: Obstructive sleep apnea following pharyngeal flap. *Cleft palate Cranio fac J*. 1997. 34; 256.
22. **Friedman M**. Palatal stiffening as a CPAP alternative. Sleep review; *Journal for sleep specialists*. Jun 2007. (sleepeditor@ascendmedia.com).

23. **Cistulli P**, Palmisano RG, Poole MD. Treatment of obstructive sleep apnea syndrome by rapid maxillary expansion. *Sleep*. 1998. 21; 831-835.

24. **Hartgerink DU**, Vig PS, Abbot DW. The effect of rapid maxillary expansion on nasal resistance. *Am J Orthod Dentofacial Orthop*. 1987.92(2). 381-389.

25. **Babacan H**, Sukucu O, Doruk C, Sinan A . Rapid maxillary expansion and surgically assisted rapid maxillary expansion effects on nasal volume.*The Angle Orthodontist*. 2006. 76(1). 66-71.

26. **Chin M**, Toth BA: Distraction osteogenesis in maxillofacial surgery using internal devices : a review of five cases. *Int J Oral Maxillofac Surg,* 1996. 54; 45 – 53.

27. **Spier S,** Rivlin J, Rowe RD, Egan T. Sleep in Pierre –Robin syndrome. *Chest*. 1986. 90; 711-715.

28. **Johnson C**, Taussing LN, Koopman C et al. Obstructive sleep apnea in Treacher Collin syndrome. *Cleft Palate Craniofac J.* 1985. 18. 39-43.

29. **Steinbacher DM**, Kaban LB, Troulis MJ. Mandibular advancement by distraction osteogenesis for tracheostomy children with severe micrognathia. *J Oral Maxillofac Surg*. 2005. 63 (8);1072-1079.