Original Article - Retrospective Study

Controlling the vector of distraction osteogenesis in the management of obstructive sleep apnea

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ABSTRACT

Background: Obstructive sleep apnea (OSA) in individuals with craniofacial anomalies can compromise airway and is a serious life-threatening condition. In many cases, tracheostomy is carried out as the treatment of choice. Distraction osteogenesis of the mandible as a treatment modality for OSA is very useful and may spare the need for tracheostomy or allow decannulation, yet controlling the vector of distraction is still a major challenge. We present a method for controlling the vector of distraction. **Materials and Methods:** Eight patients with severe respiratory distress secondary to a micrognathic mandible were treated by mandibular distraction osteogenesis using either external or internal devices. Temporary anchorage devices (TADs) and orthodontic elastics were used to control the vector of distraction. Cephalometric X-rays, computed tomography, and polysomnographic sleep studies were used to analyze the results. **Results:** A mean distraction of 22 mm using the internal devices and a mean of 30 mm using the external devices were achieved. Increase in the pharyngeal airway and hyoid bone advancement was also observed. Anterior-posterior advancement of the mandible was noted with no clockwise rotation. Most importantly, clinical improvement in symptoms of OSA, respiratory distress, and feeding was noted. **Conclusions:** We describe a method for controlling the vector of distraction used as a treatment for OSA. In these cases, TADs were used as an anchorage unit to control the vector of distraction. Our results show excellent clinical and radiographical results. TADs are a simple and nonexpensive method to control the vector of distraction.

Keywords: Control, distraction osteogenesis, obstructive sleep apnea, temporary anchorage devices, vector

INTRODUCTION

Obstructive sleep apnea (OSA) is a sleep disorder estimated to affect 3%–7% of the general population.[1] OSA episode is defined as the absence of breathing for 10 or more seconds despite the effort to breathe.[2] OSA is defined by the occurrence of daytime sleepiness, loud snoring, witnessed breathing interruptions, or awaking due to gasping or choking in the presence of at least five obstructive respiratory events (apnea, hypopneas, or respiratory effort-related arousals) per hour of sleep. The presence of at least 15 obstructive respiratory events per hour of sleep without symptoms is also sufficient for the diagnosis.[3] OSA results in significant impairment in daytime functioning, including excessive sleepiness, fatigue, and mood problems.[4] Compromised airway is a serious life-threatening condition and can occur commonly in individuals with craniofacial anomalies associated with micrognathia such as Pierre Robin sequence, hemifacial microsomia, Treacher-Collins and Nager syndromes.[5,6] In these disorders, the reduced size of the mandible and its retruded position can cause a posterior collapse of the tongue that may

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lead to upper airway obstruction.[7-9] In many cases, tracheostomy is carried out as the treatment of choice although it is associated with frequent morbidity.[10-12]

Distraction osteogenesis is a very useful solution in a number of pathological conditions such as alveolar bone deficiency for restoration purposes,[13,14] compromised airway,[15-18] and alveolar clefts.[19]

One of the main challenges during the distraction process is controlling the direction of the newly formed bone. In cases of mandible elongation as a treatment for OSA, the vector of lengthening should be forwarded to advance the mandible and hyoid bone, thus enlarging the airway and inspiratory airflow. During the mandibular distraction, an inferior component of the vector reduces the effect of forward mandibular advancement, thus making the distraction process less effective. In addition, the distraction of the mandible without controlling the vector can cause asymmetry or a clockwise rotation of the mandible resulting in an open bite, improper occlusion, and less effective forward traction of the mandible [Figure 1c].[20]

For these reasons, controlling the forward advancement of the mandible is an important principal when using distraction osteogenesis as a treatment for improvement of airway obstruction.

The distraction of the mandible can be performed by extraoral or intraoral devices. When using extraoral devices, there are unidirectional[15,16] or multidirectional, multiplanar[20] distraction devices which assist in controlling the vector of distraction.

When using internal devices, controlling the forward vector of distraction is more challenging. To assist in controlling the forward vector of distraction in internal devices, internal curvilinear distracters were introduced.[21,22] Although there was a great improvement in controlling the forward vector of distraction by external multidirectional and the internal curvilinear, there are still difficulties in controlling the forward vector of the mandible and occlusal adjustment.

When trying to control the vector of creation of newly formed bone, one can use teeth, if exist, as anchorage to maintain a desirable vector. This approach compromises existing teeth and can result in movement and rotation.[23] In addition, when treating children in their first 2–3 years of life, using teeth is not an option.

Temporary anchorage devices (TADs) are fixed temporarily to the bone and are removed after usage; their initial purpose was orthodontic anchorage, supporting the teeth of the reactive unit or replacing the reactive unit altogether.[24-26] TADs are small in size, low in cost, include minimal anatomic limitations for placement, require only a simple procedure for insertion, and can bear immediate loading.

Recently, the usage of TADs as a new alternative for controlling the vector of distraction has developed. We previously described the usage of TADs to control the vector of distraction for restoration purpose[27] and for alveolus deficiency due to cleft palate.[28] TADs were also used for controlling midface advancement through Le Fort III and monobloc distraction osteogenesis.[29]

In this manuscript, we describe a technique for controlling the vector of distraction using TADs when treating patients with OSA due to craniofacial anomalies.

MATERIALS AND METHODS

Eight patients aged from 5 months to 3 years with OSA secondary to a micrognathic mandible associated with craniofacial anomalies as Pierre Robin sequence, Goldenhar syndrome, and Treacher-Collins syndrome were treated.

The patients suffered from difficulty in breathing and eating since birth, and some were fed through a percutaneous endoscopic gastrostomy tube.

Polysomnographic sleep studies revealed respiratory disturbance index (RDI) >10 apneas per hour in all patients and oxygen saturation of $<85\%$ in all patients.

The operations were done under general anesthesia. Two different distraction devices were used; half of the patients were introduced with extraoral distraction devices and half with intraoral devices.

The lateral surface of the mandibular body was exposed between the mental nerve anteriorly and the gonial angle posteriorly. An osteotomy was performed in the mandibular body just anterior to the gonial angle [Figure 1a] and two distraction devices were mounted, one on each side of the mandible [Figure 1b]. TADs

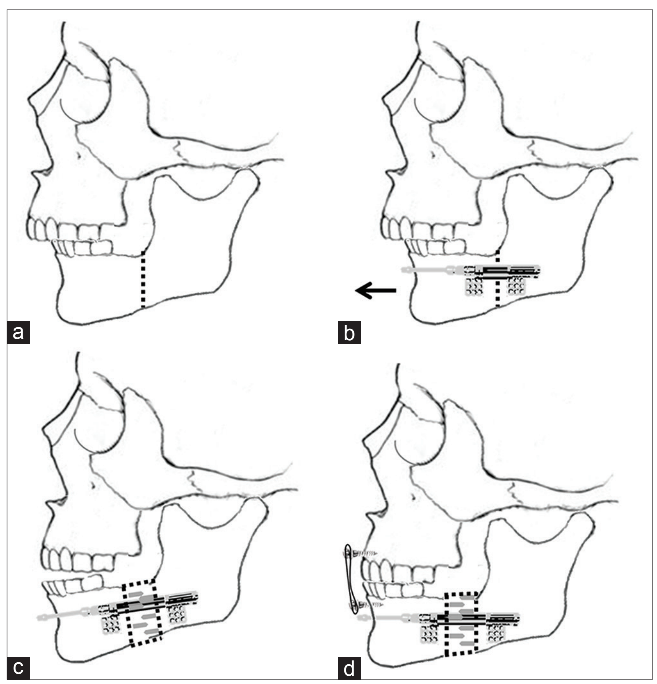

Figure 1: An illustration of a mandible distractor used for mandibular forward lengthening. (a) An osteotomy is performed as demonstrated by the dashed line anterior to the gonial angle. (b) The distractor is fitted on both fragments. (c) Bone elongation of the mandibular body is performed gradually without control over the vector of distraction; thus, a marked downward vector is observed. Dashed square and gray rods demonstrate the newly formed bone. (d) Placement of temporary anchorage devices in the anterior part of the mandible and maxilla. Elastics are used to maintain the forward vector and a correct occlusion during elongation

were placed by self-tapping, two in the anterior alveolus maxilla, and two in the anterior alveolus mandible. Great care was taken to minimize damage to the dental roots/dental buds.

Due to difficult intubation, some of the patients were left intubated for an additional 24 h under monitoring in the pediatric intensive care unit until the swelling was reduced.

Following a latency period of 3 days for primary callus organization, a gradual lengthening of the mandible was performed bilateral by activation of the distraction devices at a rate of 1 mm/day [Table 1].

During the active lengthening of the mandible, orthodontic elastics were connected to the TADs to control the forward vector of the distraction of the mandible and minimize the downward vector [Figure 1d].

To enable bone mineralization with minimal load bearing and thus minimize relapse, the devices were left for three additional months before removal after active distraction was finished.

Cephalometric X-rays and computed tomography (CT) in sagittal and axial planes were used to measure changes in airway and mandibular advancement.

Patients underwent polysomnographic sleep studies prior and postdistraction to measure improvement in physiological activity.

Clinical photographs were taken to observe soft tissue changes.

RESULTS

Cephalometric analysis demonstrated hyoid bone advancement along the axis of the mandibular body after a mean distraction of 22 mm using the internal devices and a mean of 30 mm using the external devices. The increase in pharyngeal airway was also observed.

Anterior-posterior advancement of the mandible was noted with no clockwise rotation.

Newly formed bone was well observed clinically and radiographically.

Following distraction, there was marked clinical improvement in symptoms of OSA, respiratory distress, and feeding.

Polysomnographic sleep studies revealed RDI >10 apneas/h and oxygen saturation of <85% before the distraction period in all patients and an RDI of <3 episodes/h and oxygen saturation larger than 95% postdistraction for all of the patients.

The mean change in posterior airway space as measured at C2 level in all eight patients was 2 mm preoperatively to 12 mm postoperatively.

An example of a cephalometric radiograph of the result of mandibular distraction in a 16-month-old baby suffering from Pierre Robin is shown in Figure 2. One can observe the TADs in the alveolar bone of the maxilla and the mandible.

In the same patient, one can observe the significant improvement in airway due to the distraction of the mandible as measured at the level of C2 in a CT scan as shown in Figures 3 and 4, sagittal and axial reconstructions accordingly. Three-dimensional airway volume analysis of the same patient is shown in Figure 5, an airway change from 1764 mm^2 to 12,785 mm^2 can be observed. Intraoperative photographs are shown in Figure 6, including TADs placement. The clinical result of the mandibular distraction osteogenesis is shown in Figure 7, including 2-year follow-up.

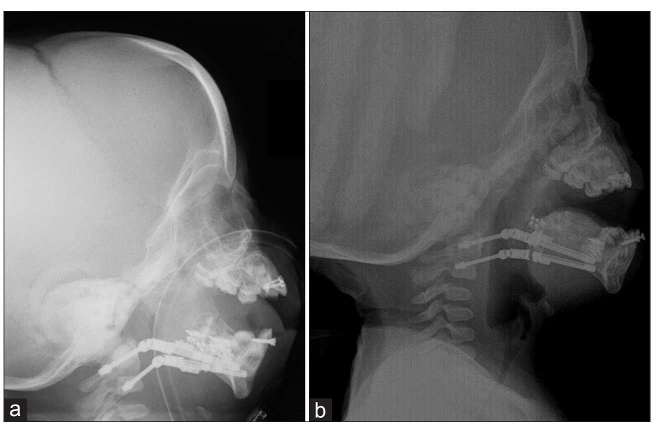

Figure 2: Lateral cephalometric radiograph showing the result of the distraction osteogenesis on the mandible and the airway. (a) Postoperative radiograph, before the initiation of distraction. (b) Radiograph taken after completion of the distraction and before the removal of the distraction device

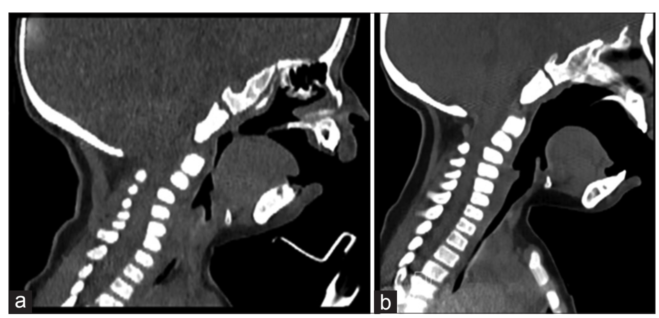

Figure 3: Sagittal reconstruction of computed tomography showing the result of the distraction osteogenesis on the location of the mandible and the expansion of the airway. (a) Preoperative computed tomography, before the initiation of distraction. (b) Radiograph taken after completion of the distraction and removal of the distraction devices

Table 1: Protocol for gradual distraction of the mandible as treatment for obstructive sleep apnea

Surgery	Latency period	Rate of bone elongation	Consolidation period	Device removal	Decannulation in case tracheostomy was performed
	3 days	1 mm/day as necessary	3 months		

Figure 4: Axial reconstruction of computed tomography showing the result of the distraction osteogenesis on the expansion of the airway. (a) Preoperative computed tomography, before the initiation of distraction. (b) Radiograph taken after completion of the distraction and removal of the distraction devices

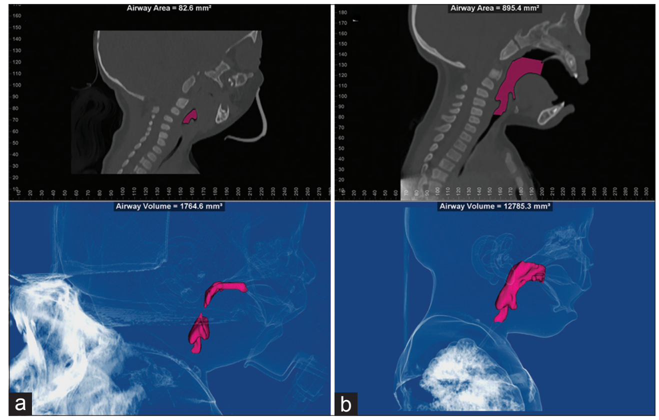

Figure 5: Airway volume analysis using (Dolphin Imaging Systems, Chatsworth, CA, USA). Airway volume of the oropharynx as measured between the caudal point of the hyoid up to the hard palate; (a) predistraction, (b) postdistraction. Significant change in airway volume is observed

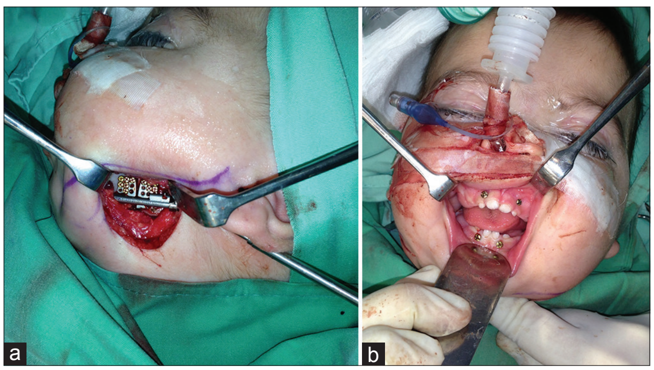

Figure 6: Intraoperative photographs of one of the patients. (a) Lateral view following osteotomy and device placement. (b) Anterior view showing the four temporary anchorage devices placed in the anterior region of the maxilla and mandible

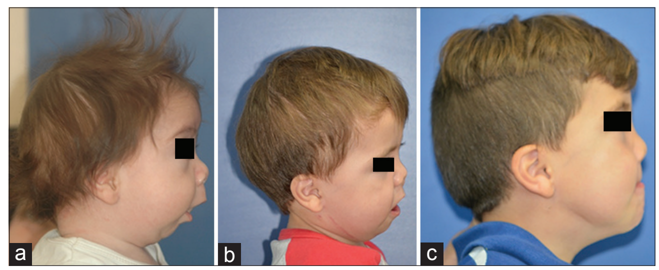

Figure 7: Profile clinical shot showing the result of the distraction osteogenesis on the mandible. (a) Preoperative photograph, before the initiation of distraction. (b) Postoperative photograph taken after completion of the distraction and removal of the distraction devices. (c) Two years following distraction osteogenesis, anterior position of the mandible with no significant vertical lengthening can be observed

DISCUSSION

There are many causes for OSA. Regardless of the underlying cause, it is the resulting disproportion in skeletal and soft-tissue dimensions that anatomically compromises the upper airway.[30]

Classically, adenotonsillectomy has been the surgical treatment of choice for pediatric OSA. However, many children, especially those with underlying medical conditions such as Down syndrome or craniofacial anomalies, require further treatment after this surgery.[31]

Long-standing tracheostomies are associated with high morbidities such as tracheomalacia, chronic bronchitis, laryngeal stenosis, and risk of death due to mucus plug or dislodgement of the tracheostomy tube. Permanent tracheostomy significantly increases the level of home or institutional care and may lead to deleterious psychological, communicative, and social effects in the affected child.[7,11,32-34]

Mandibular advancement by distraction is a useful approach that may spare the need for tracheostomy in individuals with craniofacial anomalies suffering from OSA or to allow a decannulation in severe cases with a permanent tracheostomy.[9,35-41]

The location of the osteotomy performed was located in the mandibular body, just anterior to the gonial angle to elongate the mandible, pull the suprahyoid muscles, base of tongue, and hyoid bone forward and minimize the vertical vector, thus maximizing the enlargement of the pharyngeal airway.

Even when the location of the osteotomy is ideal, the vertical component of the vector still exists due to masticatory forces. For this reason, one of the main challenges during the distraction process remains controlling the forward direction of the mandibular movement.

When trying to control the vector of creation of newly formed bone, one can use teeth, if exist, but this approach compromises existing teeth. The patients in this study were younger than 3 years old; at this age, using teeth as an anchorage for orthodontic elastics to control the vector of distraction was not an option.

We describe here a method for controlling the vector of distraction used as a treatment for OSA. In these cases, TADs were used as an anchorage unit to control the vector of distraction. Orthodontic elastics were used to connect the TADs in the mandible and maxilla, thus minimizing the vertical component of the vector.

Our results show excellent clinical and radiographical results. A significant improvement in respiratory distress and feeding was observed as well as evidential advancement of the mandible.

CONCLUSION

Using TADs for controlling the vector of distraction is a simple, none-expensive, minimally time-consuming method. TADs is a useful technique for controlling the vector of mandibular distraction in OSA patients, thus maximizing the forward vector and minimizing the undesired vertical vector, resulting in efficient airway enlargement.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Punjabi NM. The epidemiology of adult obstructive sleep apnea. Proc Am Thorac Soc 2008;5:136-43.
2. Kushida CA, Littner MR, Morgenthaler T, Alessi CA, Bailey D, Coleman J Jr., *et al.* Practice parameters for the indications for polysomnography and related procedures: An update for 2005. Sleep 2005;28:499-521.
3. Epstein LJ, Kristo D, Strollo PJ Jr., Friedman N, Malhotra A, Patil SP, *et al.* Clinical guideline for the evaluation, management and long-term care of obstructive sleep apnea in adults. J Clin Sleep Med 2009;5:263-76.
4. Aloia MS, Arnedt JT, Davis JD, Riggs RL, Byrd D. Neuropsychological sequelae of obstructive sleep apnea-hypopnea syndrome: A critical review. J Int Neuropsychol Soc 2004;10:772-85.
5. Cosman B, Crikelair GF. Mandibular hypoplasia and the late development of glossopharyngeal airway obstruction. Plast Reconstr Surg 1972;50:573-9.
6. Sjolin S. Hypoplasia of the mandible as a cause of respiratory difficulties in the infant. Acta Paediatr 1950;39:255-61.
7. Cohen SR, Simms C, Burstein FD. Mandibular distraction osteogenesis in the treatment of upper airway obstruction in children with craniofacial deformities. Plast Reconstr Surg 1998;101:312-8.
8. Perkins JA, Sie KC, Milczuk H, Richardson MA. Airway management in children with craniofacial anomalies. Cleft Palate Craniofac J 1997;34:135-40.
9. Williams JK, Maull D, Grayson BH, Longaker MT, McCarthy JG. Early decannulation with bilateral mandibular distraction for tracheostomy-dependent patients. Plast Reconstr Surg 1999;103:48-57.
10. Cohen SR, Simms C, Burstein FD, Thomsen J. Alternatives to tracheostomy in infants and children with obstructive sleep apnea. J Pediatr Surg 1999;34:182-6.
11. Guilleminault C. Obstructive sleep apnea syndrome and its treatment in children: Areas of agreement and controversy. Pediatr Pulmonol 1987;3:429-36.
12. Sidman JD, Sampson D, Templeton B. Distraction osteogenesis of the mandible for airway obstruction in children. Laryngoscope 2001;111:1137-46.
13. Laster Z, Rachmiel A, Jensen OT. Alveolar width distraction osteogenesis for early implant placement. J Oral Maxillofac Surg 2005;63:1724-30.
14. Uribe F, Agarwal S, Janakiraman N, Shafer D, Nanda R. Bidimensional dentoalveolar distraction osteogenesis for treatment efficiency. Am J Orthod Dentofacial Orthop 2013;144:290-8.
15. Rachmiel A, Aizenbud D, Pillar G, Srouji S, Peled M. Bilateral mandibular distraction for patients with compromised airway analyzed by three-dimensional CT. Int J Oral Maxillofac Surg 2005;34:9-18.
16. Rachmiel A, Emodi O, Aizenbud D. Management of obstructive sleep apnea in pediatric craniofacial anomalies. Ann Maxillofac Surg 2012;2:111-5.
17. Hamada T, Ono T, Otsuka R, Honda E, Harada K, Kurabayashi T, *et al.* Mandibular distraction osteogenesis in a skeletal class II patient with obstructive sleep apnea. Am J Orthod Dentofacial Orthop 2007;131:415-25.
18. Conley RS, Legan HL. Correction of severe obstructive sleep apnea with bimaxillary transverse distraction osteogenesis and maxillomandibular advancement. Am J Orthod Dentofacial Orthop 2006;129:283-92.
19. Rachmiel A, Even-Almos M, Aizenbud D. Treatment of maxillary cleft palate: Distraction osteogenesis vs. orthognathic surgery. Ann Maxillofac Surg 2012;2:127-30.
20. McCarthy JG, Williams JK, Grayson BH, Crombie JS. Controlled multiplanar distraction of the mandible: Device development and clinical application. J Craniofac Surg 1998;9:322-9.
21. Kaban LB, Seldin EB, Kikinis R, Yeshwant K, Padwa BL, Troulis MJ. Clinical application of curvilinear distraction osteogenesis for correction of mandibular deformities. J Oral Maxillofac Surg 2009;67:996-1008.
22. Miller JJ, Kahn D, Lorenz HP, Schendel SA. Infant mandibular distraction with an internal curvilinear device. J Craniofac Surg 2007;18:1403-7.
23. Herford AS, Audia F. Maintaining vector control during alveolar distraction osteogenesis: A technical note. Int J Oral Maxillofac Implants 2004;19:758-62.
24. Fritz U, Ehmer A, Diedrich P. Clinical suitability of titanium microscrews for orthodontic anchorage-preliminary experiences. J Orofac Orthop 2004;65:410-8.
25. Kanomi R. Mini-implant for orthodontic anchorage. J Clin Orthod 1997;31:763-7.
26. Miyawaki S, Koyama I, Inoue M, Mishima K, Sugahara T, Takano-Yamamoto T. Factors associated with the stability of titanium screws placed in the posterior region for orthodontic anchorage. Am J Orthod Dentofacial Orthop 2003;124:373-8.
27. Aizenbud D, Hazan-Molina H, Cohen M, Rachmiel A. Combined orthodontic temporary anchorage devices and surgical management of the alveolar ridge augmentation using distraction osteogenesis. J Oral Maxillofac Surg 2012;70:1815-26.
28. Rachmiel A, Emodi O, Gutmacher Z, Blumenfeld I, Aizenbud D. Oral and dental restoration of wide alveolar cleft using distraction osteogenesis and temporary anchorage devices. J Craniomaxillofac Surg 2013;41:728-34.
29. Francis C, Rommer E, Mancho S, Carey J, Hammoudeh JA, Urata MM. Vector control in internal midface distraction using temporary anchorage devices. J Craniofac Surg 2012;23 7 Suppl 1:2000-3.
30. Cohen SR, Ross DA, Burstein FD, Lefaivre JF, Riski JE, Simms C. Skeletal expansion combined with soft-tissue reduction in the treatment of obstructive sleep apnea in children: Physiologic results. Otolaryngol Head Neck Surg 1998;119:476-85.
31. Marcus CL. Management of obstructive sleep apnea in childhood. Curr Opin Pulm Med 1997;3:464-9.
32. Argamaso RV. Glossopexy for upper airway obstruction in Robin sequence. Cleft Palate Craniofac J 1992;29:232-8.
33. Conway WA, Victor LD, Magilligan DJ Jr., Fujita S, Zorick FJ, Roth T. Adverse effects of tracheostomy for sleep apnea. JAMA 1981;246:347-50.
34. Sasaki CT, Horiuchi M, Koss N. Tracheostomy-related subglottic stenosis: Bacteriologic pathogenesis. Laryngoscope 1979;89(6 Pt 1):857-65.
35. Denny AD, Talisman R, Hanson PR, Recinos RF. Mandibular distraction osteogenesis in very young patients to correct airway obstruction. Plast Reconstr Surg 2001;108:302-11.
36. Iatrou I, Theologie-Lygidakis N, Schoinohoriti O. Mandibular distraction osteogenesis for severe airway obstruction in Robin sequence. Case report. J Craniomaxillofac Surg 2010;38:431-5.
37. Miloro M. Mandibular distraction osteogenesis for pediatric airway management. J Oral Maxillofac Surg 2010;68:1512-23.
38. Monasterio FO, Drucker M, Molina F, Ysunza A. Distraction osteogenesis in Pierre Robin sequence and related respiratory problems in children. J Craniofac Surg 2002;13:79-83.
39. Morovic CG, Monasterio L. Distraction osteogenesis for obstructive apneas in patients with congenital craniofacial malformations. Plast Reconstr Surg 2000;105:2324-30.
40. Nørholt SE, Jensen J, Schou S, Pedersen TK. Complications after mandibular distraction osteogenesis: A retrospective study of 131 patients. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2011;111:420-7.
41. Rachmiel A, Emodi O, Rachmiel D, Aizenbud D. Internal mandibular distraction to relieve airway obstruction in children with severe micrognathia. Int J Oral Maxillofac Surg 2014;43:1176-81.