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Leading Clinical Paper
Distraction Osteogenesis

Bilateral mandibular distraction for patients with compromised airway analyzed by three-dimensional CT

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Abstract. The purpose of this study was to present the method of mandibular distraction osteogenesis in order to improve airway to respiratory distressed patients due to significant mandibular deficiency, and to present the quantitative volumetric evaluation of mandible and upper airway using three-dimensional-CT (3D-CT) before and after distraction.

This study involved 12 patients aged 12 months to seven years with various complaints of Obstructive Sleep Apnea (OSA) such as noisy breathing during sleep, waking episodes, pauses in respiration and daytime somnolence. Some of them were considered tracheostomy candidates. All the patients underwent bilateral mandibular distraction under general anesthesia. 3D-CT of face and neck was performed before and after distraction and a quantitative volumetric evaluation of mandibular volume and airway volume was performed.

The results reveal successful mandibular advancement with increase of mandibular volume by an average of 28.24% and increase of upper airway volume with a mean of 71.92%. Moreover, there were improved apnea index and oxygen saturation and elimination of OSA symptoms. In conclusion, the results demonstrate that following distraction osteogenesis of hypoplastic mandible the volume of the mandible and upper airway increases, eliminating symptoms of OSA and preventing tracheostomy.

Key words: mandibular distraction osteogenesis; obstructive sleep apnea; upper airway.

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Upper airway obstruction occurs most commonly in individuals with craniofacial anomalies associated with micrognathia as Pierre Robin syndrome, Hemifacial Microsomia, Treacher Collins and Nager syndromes[5,26]. In these disorders the reduced size of mandible and its retruded position cause retro displacement of the tongue and concomitant reduction of the oropharyngeal airway that may lead to upper airway obstruction. The patients have symptoms of Obstructive Sleep Apnea (OSA) that in severe cases need tracheal intubation and tracheostomy[4,19,27].

Distraction osteogenesis has become an accepted method of treatment for patients requiring reconstruction of hypoplastic mandibles and may achieve mandibular lengthening without need for

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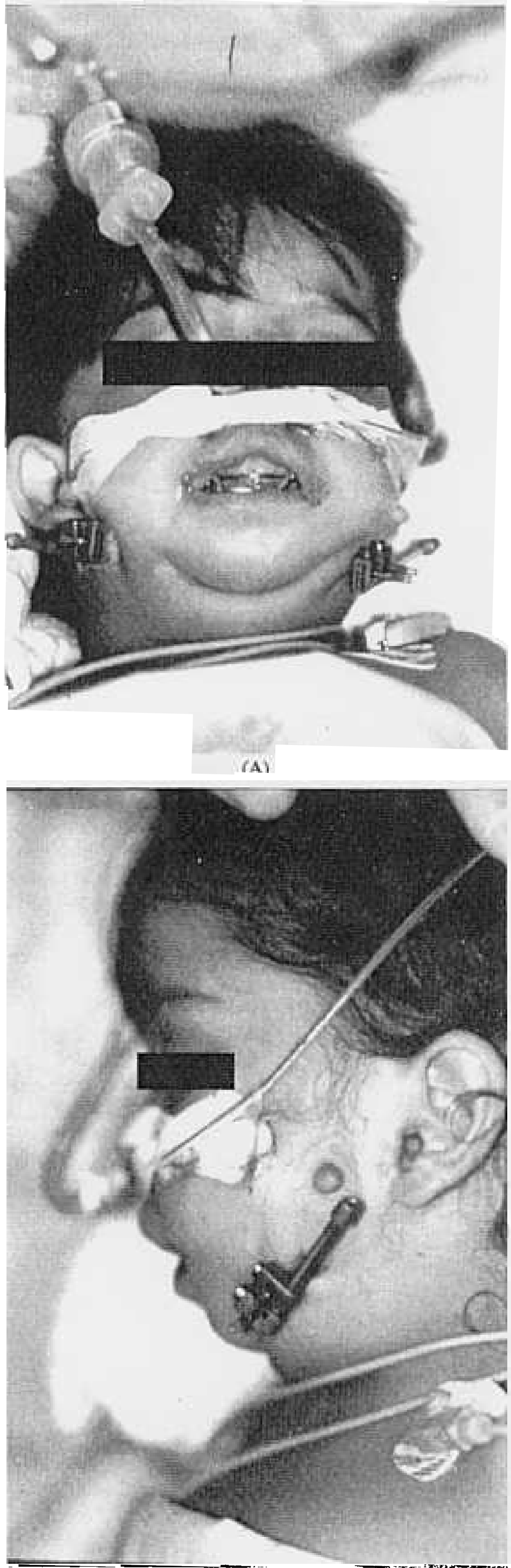
(A)

(B)

Fig. 1. Two and a half-year-old girl with Goldenhar syndrome and obstructive sleep apnea due to hypoplastic mandible. Preoperative facial photographs. (A) AP, (B) lateral.



(A)

(B)

Fig. 2. The patient at the end of operation, before commencement of distraction. (A) AP, (B) lateral.

bone graft[22,23]. During the past few years mandibular reconstruction by distraction osteogenesis has been demonstrated as effective in resolving the upper airway obstruction and for tracheostomy decannulation[4,17,18,27]. Mandibular distraction has also been used in respiratorily distressed neonates and infants to avoid tracheostomy[8,18].

A computerized tomographic (CT) scan technique for quantitative volumetric assessment of mandible after distraction osteogenesis (DO) was proved to be an accurate method to measure the amount of bone regeneration[25]. Using this technique an increase in the volume of the mandible and the upper airway in children with mandibular micrognathia following distraction osteogenesis has been shown[20].

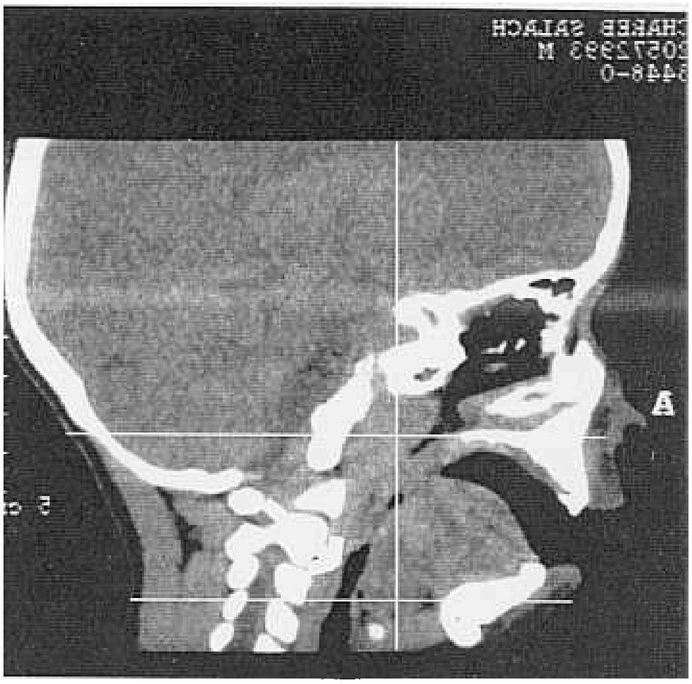
The purpose of this study is to present the quantitative volumetric evaluation of mandibular and upper airway size in young patients having distraction osteogenesis for micrognathia using 3D-CT.

Materials and methods

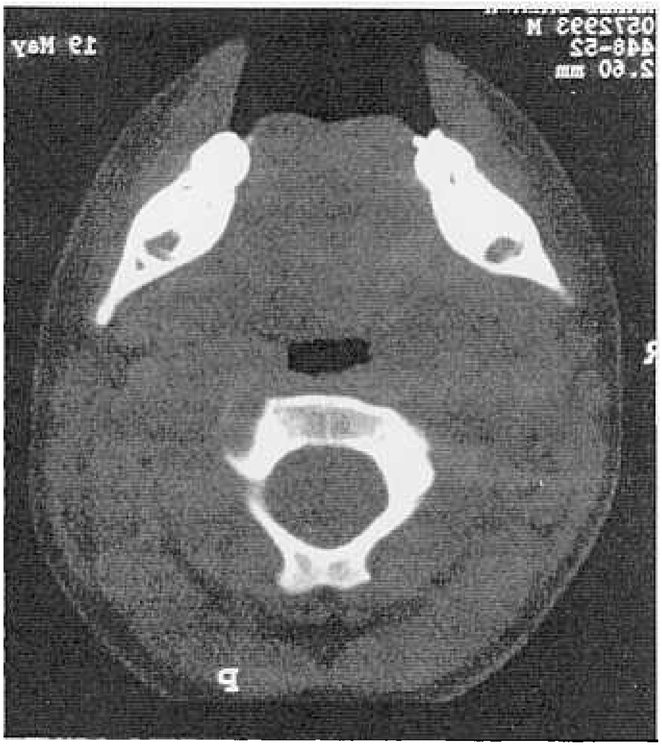
Over the past three years 12 patients (5 males and 7 females) aged 12 months–7 years, with various symptoms of OSA were treated (Fig. 1). All had mechanical upper airway obstruction that was determined by clinical and radiological evidence of severe hypoplastic mandible and symptoms of OSA as noisy breathing during sleep, fragmental sleep, pauses in respiration and daytime somnolence. In some of them tracheostomy was considered. Eight of the patients were under Continuous Positive Airway Pressure (CPAP) treatment. Polysomnographic sleep studies revealed respiratory disturbance index greater than 10 apneas per hour and oxygen saturation less than 85% (Table 1).

Each patient was evaluated before treatment and after treatment, one month after removal of distraction devices with computer tomography (CT) in axial and sagittal planes and three-dimensional CT of the facial bones and upper airway. Mandibular distraction was planned bilaterally in order to advance the mandible and to increase the upper airway volume. Panoramic and lateral cephalometry were performed before the operation, during the distraction and at the end of retention period and after removal of the devices. These X-rays were used for operative planning and follow-up but are not presented in this study.

The surgical procedure was performed under general anesthesia with nasotracheal intubation on both sides of the mandible. A muco-periosteal incision was performed intraorally on the buccal side of the mandible exposing the mandibular body from the gonial area to the mental nerve. Consequently buccal and lingual corticotomies were performed in the mandibular body including the lower and upper border of the mandible. Two extraoral percutaneous pins were



(A)



(B)

Fig. 3. Anatomic limits of upper airway in sagittal CT scan. The arrow demonstrates the airway measured between the lines (A). The axial CT scan (B).

Table 1. Polysomnography: (A) preoperative respiratory disturbance index (RDI)–apnea-hypopnea index and (B) preoperative O_2 saturation

	Respiratory evaluation					
	(A) RDI–apnea-hypopnea index			(B) Average oxygen saturation (%)		
Patient number	Pre distraction	Post 1[a]	Post 2[b]	Pre distraction	Post 1[a]	Post 2[b]
2		0				
3		2	2			
4		2	2			
5		1	2			
6		2	2			
7		2	2			
8		0				
9			2			
10		2	2			
11		2	2			
12		2	2			
Average		1.4	1.8			
SD		0.8	0.4			

(A) Preoperative respiratory disturbance index (RDI)–apnea-hypopnea index was greater than 10 episodes per hour, decreasing after removal of the device and one year later to less than 2 per hour (RDI) (t-test preoperative – post 1: $P < 1 \times 10^{-8}$). (B) Preoperative O_2 saturation was less than 85%. After removal of the device and one year later it was over 95% in all patients (t-test preoperative – post 1: $P < 1 \times 10^{-9}$).
[a] Post 1 – one month after removal of distraction devices.
[b] Post 2 – over one year after distraction.

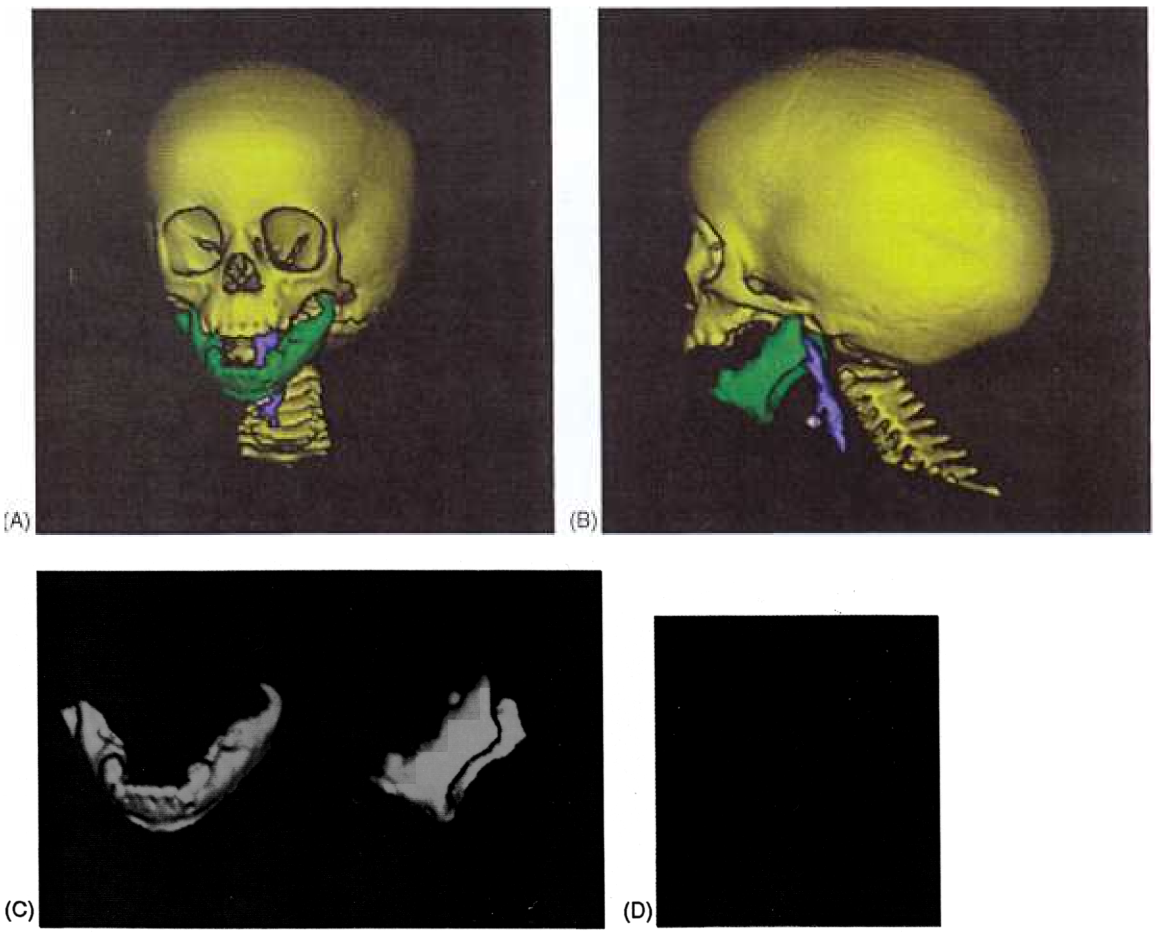


Fig. 4. AP and lateral aspect of three-dimensional digital image demonstrating pre operative hypoplastic mandible (in green) and airway (in blue) ((A) AP, (B) lateral), with isolation of the mandible (C) and airway (D) for volume quantification.

inserted bicortically, one anterior and the other posterior to the corticotomy. Upon clinical inspection and X-ray observation, great care was taken not to damage the dental roots or buds, and the inferior alveolar nerve. After pin insertion, using fine chisels on the periphery of the bone, completion of the osteotomy was performed. Upon completion of the osteotomy, a unidirectional extraoral distraction device was connected to the pins while the teeth were in the previous preoperative occlusion and the fracture made was reduced and fixed with the distraction device. Suturing of the periosteum and buccal mucosa was performed (Fig. 2).

The patients with severe OSA were maintained several days in intensive care unit for monitoring the postoperative airway and oxygen saturation till the swelling reduced and the distraction was initiated. The distraction was initiated bilaterally after four days of latency period for primary callous organization in a rate of 0.5 mm twice a day, a total of 1 mm per day between 26 and 33 mm as was necessary followed by 10 weeks of consolidation period. All the patients were treated by local disinfection daily around the pins.

Regarding computed tomographic scanning, each child received the same protocol for the three-dimensional CT of whole head, with 1 mm continuous axial slices, parallel to the Frankfurt horizontal[14]. Three-dimensional volumetric reconstruction was created from archived computed tomographic digital data for evaluation using the spiral CT 4 slice scanner (Mx8000, Phillips – software: MxView version 5). The airway was demonstrated as well. The following limits for upper airway volume were adopted (Fig. 3):

cephalad – the hard palate plane,
caudad – the glottis,
ventral – the junction of the superior adenoid tissue and the nasopharynx,
dorsal – the posterior pharyngeal wall, and
lateral – the right and left lateral pharyngeal walls.

The computed tomographic digital data were used to evaluate the upper airways and mandibular volumes pre distraction and post distraction (Fig. 4). The volumetric anatomically complete mandible was extracted using a bone soft tissue interface (Fig. 4C). Using parameters to extract only the data within the anatomic constraints, a digital set of the edited upper airway volume was obtained (Fig. 4D). The volume determination was used for volumetric qualification of both upper airway and the mandible before and after distraction. The patients were followed clinically and by polysomnography between 12 and 24 months. Polysomnography was performed to all children one month after removal of the distraction devices and again 12 months later and compared to the polysomnography before distraction.

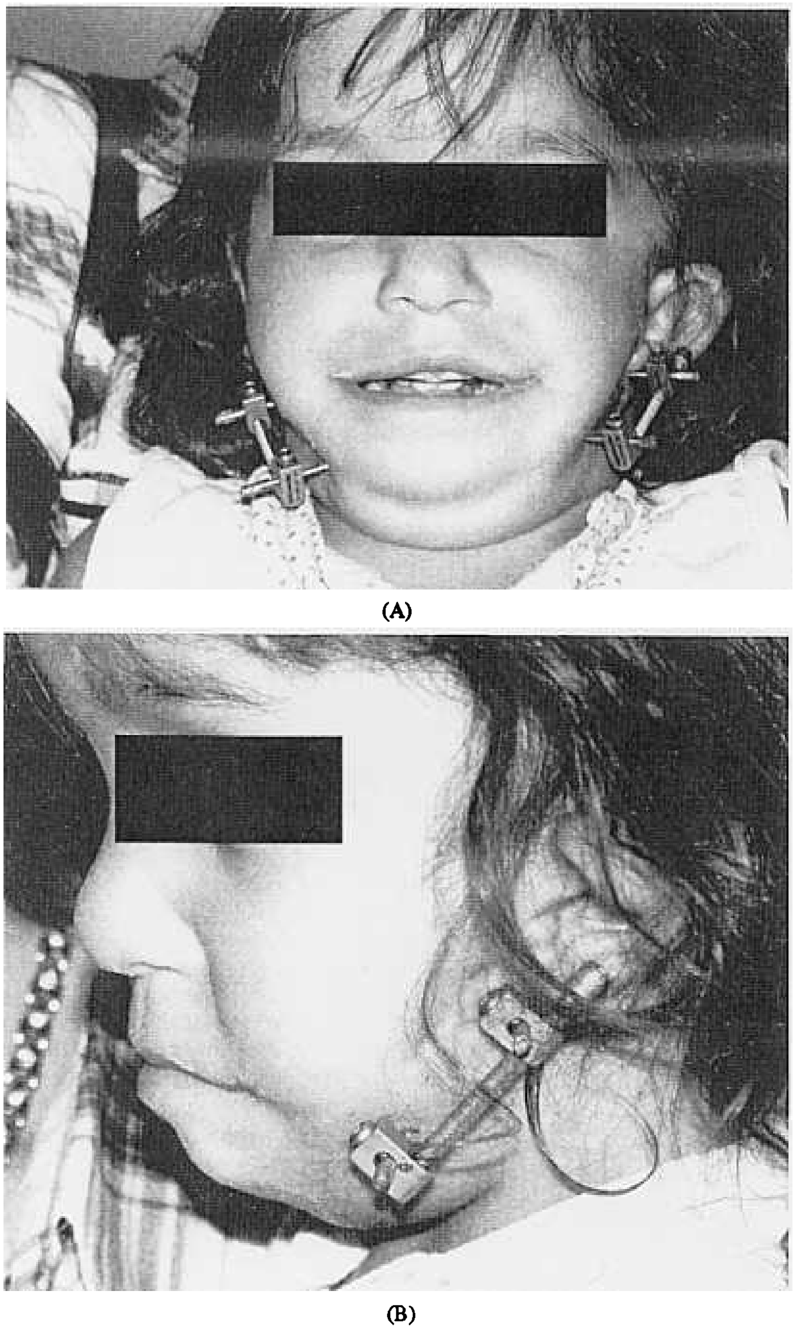


Fig. 5. The patient at the end of bilateral mandibular distraction, during the retention period. (A) AP, (B) lateral.

Results

During the first postoperative days, the patients developed postoperative edema in the submandibular area bilaterally, aggravating the compromised airway. Therefore, during the first five postoperative days children with severe air-

(A)

(B)

Fig. 6. After removal of the distraction devices and elongation of the mandible. (A) AP, (B) lateral.

way obstruction were maintained in the intensive care unit until the edema was reduced and mandibular advancement commenced. Mandibular distraction was successful in all 12 patients. There were no complications with the distraction devices and no signs of infection. The average linear mandibular lengthening measured by the device rod was 28 mm (range 25–31 mm) on the left and 29 mm (range 26–32 mm) on the right side (Figs 5 and 6). In all patients, a forward advancement of the mandible with class III dental occlusion was achieved.

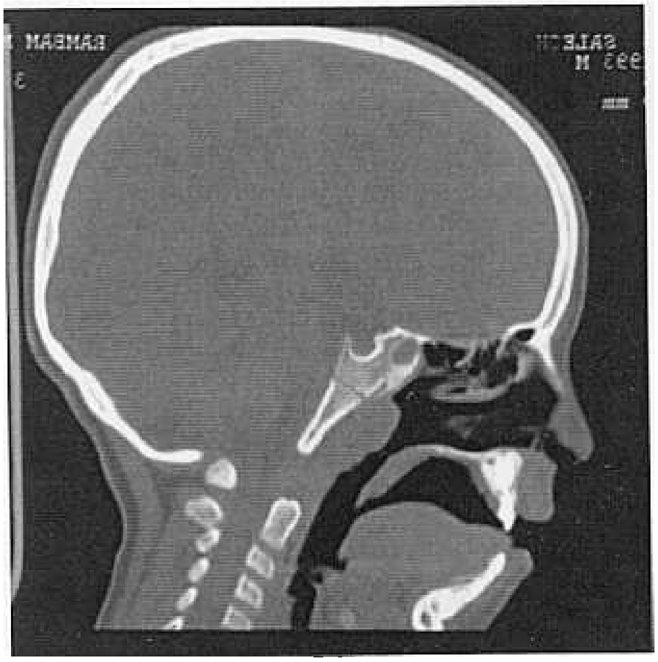
The lateral and axial CT at the end of distraction revealed forward lengthening of the mandible and hyoid bone as exemplified in Fig. 7. These resulted in forward traction of the tongue and increased pharyngeal space.

The three-dimensional digital image demonstrates the increased mandible and airway (Fig. 8). The mandibular volume increased between 23.4 and 33.4% (an average of 28.24%) (Fig. 9A). Quantitative assessment of the upper airway volume before and after distraction demonstrates increased volume ranging from 57 to 89% with a mean of 71.92% (Fig. 9B). 3D-CT demonstrated improved airway in all patients. The increase in total mandibular volume by distraction resulted in increase in upper airway volume (Fig. 10). Polysomnographic studies conducted one month after the procedure and 12 months later, showed improvement of airway obstruction in all patients (Table 1). Respiratory disturbance index of all patients was less than 2 episodes per hour and oxygen saturation gradually increased during lengthening reaching at least 95% in all patients. During the 12–24 months of follow-up none of the patients developed symptoms of OSA and none needed CPAP treatment.

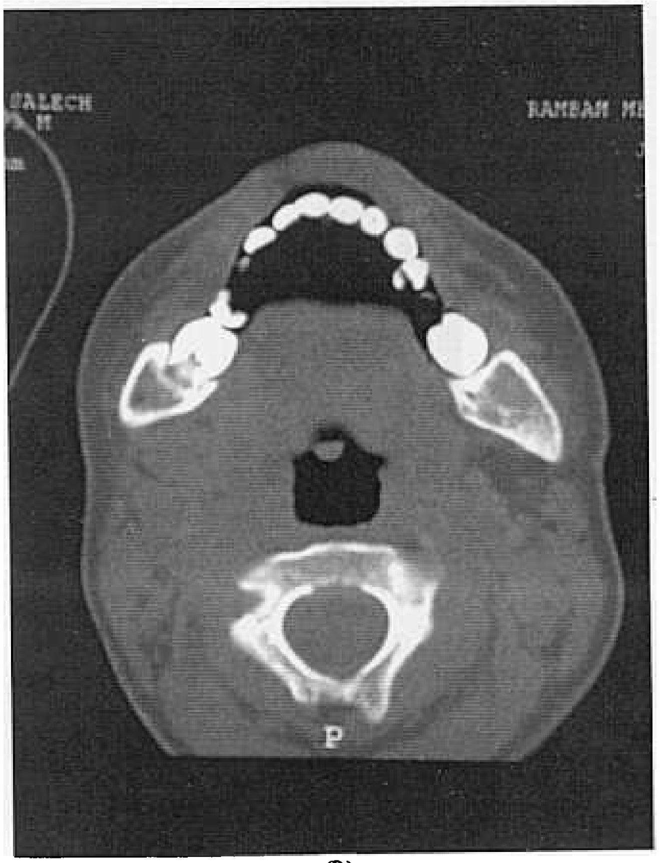
Discussion

Children with craniofacial anomalies associated with micrognathia, retruded position of the mandible and glossoptosis often have compromised upper airway, a condition with potential for morbidity and mortality[5,19,26]. In severe cases, the children are treated by permanent tracheostomy. However, when they are faced with a disturbance such as upper respiratory tract infection or palatal surgery, an obstructive respiratory condition may develop, requiring placement of tracheostomy. These patients usually fail to thrive, present feeding problems, insufficient weight gain associated with malnutrition, higher pulmonary morbidity and long-term hospitalization[4,19,26]. Long-standing tracheostomies are associated with high morbidity such as tracheomalacia, chronic bronchitis, laryngeal stenosis and risk of death due to the mucus plug or dislodgement of tracheostomy tube. Patients who undergo tracheostomy require complex nursing care[26,27].

Other procedures have been used to provide adequate airway as treatment of OSA. These include glossopexy[1] to small children in which the tongue is



(A)



(B)

Fig. 7. The post distraction sagittal slices of computed tomographic scans, showing the improvement in glossoptosis and airway obstruction (A). The axial CT demonstrates the increased airway as well (B).

moved anteriorly and attached to the mandible or lip, genioglossus advancement[15] transfixion of tongue by Kirschner Wire[11], or hyomandibulopexy[2] in which the hyoid bone and base of tongue are advanced forward. Another procedure is subperiosteal release of floor of mouth musculature[7]. The concept is that the musculature of floor of mouth is under increased tension and pushes the tongue upwards and backwards. Early release of this musculature allows the tongue to return to a more normal position. There are no published studies with objective measurements that demonstrate the benefit of this technique. Another study demonstrates that daytime submandibular electrostimulation of suprahyoidal muscles may prevent episodes of apnea in OSA[28]. Other surgical options, mainly for adults, are mandibular trapezoid osteotomy[6] or a combination of uvulopalatopharyngoplasty, mortised genioplasty and maxillomandibular advancement[12]. The method of DO has substantial advantage over all the above mentioned techniques. It enables gradual forward advancement of mandible and tongue that increase the pharyngeal space. The apnea index and O_2 saturation markedly improved following distraction.

The vector of mandibular lengthening should be forward in order to increase the mandibular frame and advance the hyoid bone. Therefore, the device should be placed parallel to the lower border of the mandible. Often, with the extraoral devices there is a tendency to open bite. Interocclusal elastics anchored on the teeth to guide the vector forward controlled this. Our results confirmed efficacy of mandibular distraction in treatment of OSA in all children, avoiding necessity for tracheostomy.

A study performed by Williams et al.[27] demonstrates expansion of mandibular framework with advancement of base of tongue that leads to increased pharyngeal airway for early decannulation of tracheostomy dependent patients. This is determined on the basis of cephalometric study measuring the advancement of hyoid bone along the axis of mandibular body after distraction. In the present study we used three-dimensional volumetric measurement of upper airway, which is more accurate in the assessment of the upper airway. In the past decade, some centers have used CT rather than cephalometric studies to assess the effect of orthognathic surgery on pharyngeal airway[13,16]. Metes et al.[16] describes the use of three-dimensional volumetric studies after orthognathic

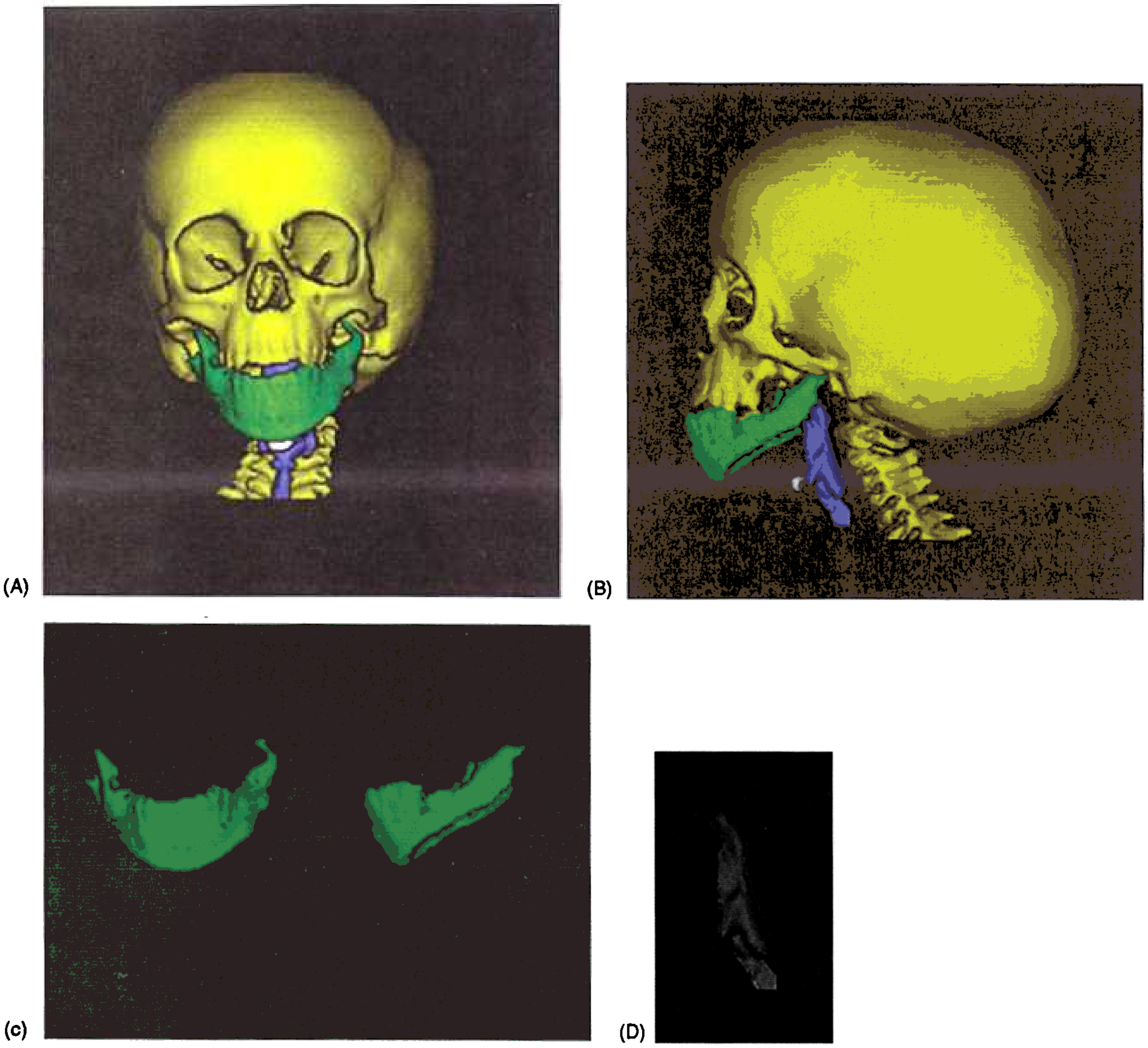


Fig. 8. AP and lateral aspect of three-dimensional digital image of facial skeleton post distraction demonstrating the increased mandible (in green) and airway (in blue) ((A) AP, (B) lateral) with isolation of the mandible (C) and airway (D) for volume quantification.

surgery in correction of OSA and Kawamata et al.[13] have used three-dimensional CT on 30 patients to assess the changes in pharyngeal airway after mandibular setback osteotomy.

Several authors have used computerized tomographic (CT) scans of craniofacial skeleton with or without image analysis software program to determine intraorbital and intracranial volumes[3,9,10,24]. Quantitative volumetric analysis of CT scans has been proved as an accurate method to measure the amount of bone regeneration in patients undergoing DO of the mandible[25]. Perlyn et al.[20], using CT scans, demonstrated increase of total mandibular volume and upper airway volume by quantitative assessment after mandibular distraction. Post processing of digital medical imaging data of computed tomographic scans, has become useful in volumetry of organs and spaces *in vivo.* The accuracy of these indirect computer measurements have been validated against direct measurements using water displacement or fill methods, with an average difference of 3.7–4.2%[21,25]. The organ planned to be measured can be rotated for full appreciation of its shape and characteristic. This method provides an alternative to the traditional lateral cephalometry for investigation of the pharyngeal space, mandibular and hyoid bone position by cephalometric landmark. Such planar radiographic images allow only two-dimensional imaging and are not as accurate as three-dimensional ones, due to overlapping shadows.

In summary, the results of this study support that distraction of the micrognathic mandible increases the volume of upper airway, increases the mandibular volume and advances the hyoid bone. There is improvement in glossoptosis and airway obstruction eliminating symptoms of OSA eliminating the need for tracheostomy.

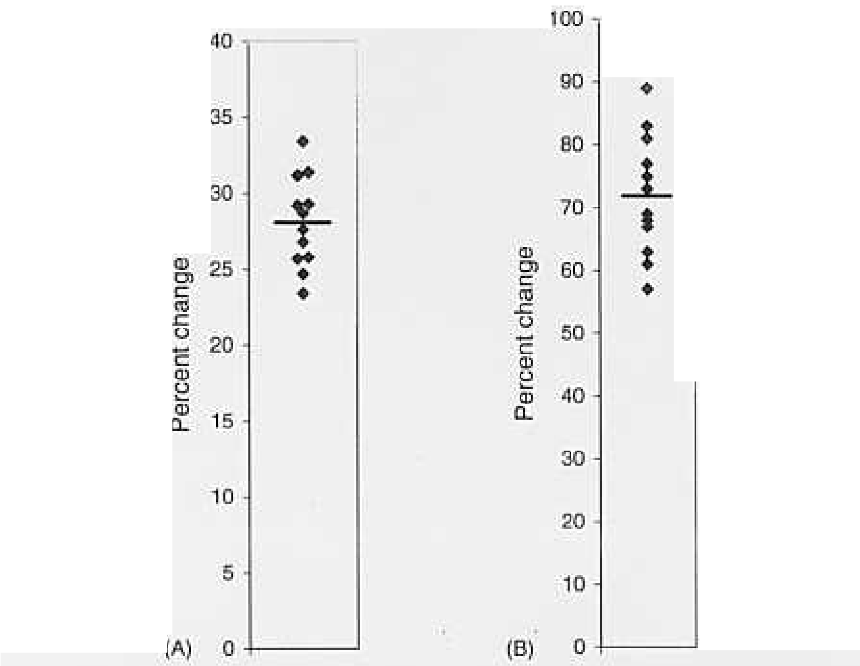


Fig. 9. Percent increase in total mandibular volume (A) and upper airway volume (B) after mandibular distraction ($n = 12$). Bars represent average value.

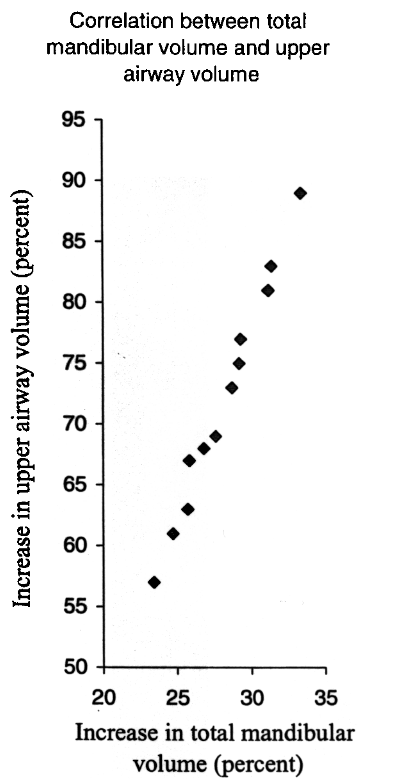


Fig. 10. Increase in total mandibular volume after distraction resulted in increase in upper airway volume.

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