Journal of Cranio-Maxillofacial Surgery (2009) **37**, 216–224

doi:10.1016/j.jcms.2008.10.014, available online at http://www.sciencedirect.com

Bilateral intra-oral distraction osteogenesis for the management of severe congenital mandibular hypoplasia in early childhood

Abdelfattah A. SADAKAH, Prof, Ph.D.[1], Mohammad A. ELSHALL, Lecturer, Assist Prof and Consultant, Ph.D.[1], Amgad A. FARHAT, Ass Prof, Ph.D.[2]

[1] *Department of Oral & Maxillofacial Surgery (Head: Prof. Mohammad S. Khedr), Faculty of Dentistry, Tanta University, Egypt;* [2] *Department of Chest, Faculty of Medicine, Tanta University, Egypt*

SUMMARY. Introduction: **Young children with severe mandibular hypoplasia usually present with varying degrees of peripheral airway obstruction and difficulty with feeding. Early treatment is important for such children. Distraction osteogenesis (DO) using intra-oral devices provides an excellent alternative when other surgical techniques do not prove to be satisfactory.** Aim of the work: **To evaluate the long-term efficacy of intra-oral bilateral DO in the treatment of severe congenital mandibular hypoplasia in early childhood.** Patients and methods: **Seven patients (4 females and 3 males), their ages ranged from 7 months to 8 years (with a mean of 34 months). They presented with severe congenital mandibular hypoplasia with obstructive sleep apnoea and difficulty in feeding. All patients were treated with bilateral mandibular DO, using an intra-oral unidirectional unburied distractor. The average follow-up period was 3.7 years (range, 2–5 years).** Results: **The patients were successfully treated using bilateral intra-oral unidirectional distractor by the use of a modified technique. After completion of distraction, retrognathia was corrected in all patients. The "subjective" symptoms had disappeared completely or had been alleviated. The mean effective airway space increase (defined by the lateral cephalograms measurements) was 70.5% (range, 31–105%, $p < 0.01$) when compared with pre-distraction. The apnoea/hypopnoea index was lowered from 60 (9.8–126.5) to 1.57 (0–16.4) and the sleep apnoea symptoms had disappeared. The mean oxygen saturation increase was from 80% to 98% post-distraction.** Conclusion: **DO can consistently produce a measurable cross-section airway improvement in patients as young as 7 months. © 2008 European Association for Cranio-Maxillofacial Surgery**

Keywords: **Distraction osteogenesis, Obstructive sleep apnoea in children, Mandibular lengthening, Surgical correction of mandibular hypoplasia**

INTRODUCTION

Sleep apnoea is one of the most frequent manifestations of respiratory obstruction. Historically, this clinical entity has stimulated the production of numerous valuable contributions with one purpose in mind: the improvement of airway resistance. A multi-disciplinary approach is required to define the problem in anatomical and functional terms to avoid a tracheostomy and prevent long-term sequelae (*Monasterio* et al., 2002).

Apnoea has been defined as cessation of airflow at the level of the nostrils and mouth lasting at least 10 s. Sleep apnoea syndrome is diagnosed if there are 5 episodes or more of apnoea per sleep hour (*Bailey* and *Croft*, 1987).

Abnormal breathing during sleep may frequently occur in early childhood, but it is usually unnoticed because of unperceived medical or physiologic sequelae. Infants and children with neuromuscular disease exhibit a higher incidence of obstructive sleep apnoea (OSA) secondary to hypotonicity of the pharyngeal musculature that allows collapse of the airway (*Cohen* et al., 1997).

In addition, the potential for airway obstruction is further increased by supine positioning, neck flexion, and increased secretion during sleep (*Hochban* and *Hoch*, 1998).

The great difficulty in diagnosing sleep apnoea is that there may be few, if any, physical signs during the day. However, in the child, a poor nasal airway with constant mouth breathing is usual. The upper lip is coated with nasal mucous and daytime respiration may be noisy variable stertor present. The child may be undersized for age and there is some evidence that chronic respiratory obstruction is largely responsible for this delay in general development, together with real physical problems in swallowing (*Williams*, 1994).

Initial management of OSA in children has traditionally included pharmacologic treatment, the use of nasopharyngeal airway devices, oxygen supplementation and positive airway pressure (*Cohen* et al., 1998).

When these fail, surgical procedures such as tonsillectomy, adenoidectomy, uvulopalatopharyngoplasty, or tracheostomy may be required to correct or bypass the abnormal anatomical features contributing to obstruction of the airway. Recent literature has suggested that facial skeletal advancement using distraction osteogenesis (DO) might be beneficial in those children with OSA secondary to midface hypoplasia or retromicrognathia and lack of tongue support (*Burstein* et al., 1995; *Rachmiel* et al., 2005).

Children born with a diminutive mandible present a challenge to paediatricians and paediatric specialists, because of the potential for airway obstruction (*Schaefer* et al., 2004).

Patients with congenital retrognathia and glossoptosis, with or without cleft palate, are correctly diagnosed with Pierre Robin sequence (*Caouette-Laberge* et al., 1994). This group represents the majority of children who are born with a combination of retrognathia and micrognathia and who may develop associated symptoms of respiratory distress (*Bear* and *Priest*, 1980). The remainders are children with mandibulofacial dysostosis disorders such as Treacher Collins syndrome, Nager syndrome, and bilateral hemifacial microsomia (*Moore* et al., 1994).

From the time that these syndromes were first described, various corrections for infant airway obstruction and the resulting condition of failure to thrive have been proposed, depending on the severity of clinical presentation. Several approaches have been used to alleviate the respiratory obstructive symptoms, which occur while awake and during feeding. The continuous prone position has historically been recommended to control mild airway obstruction. Tongue/lip adhesion procedures or tracheostomies are most commonly used to control moderate-to-severe obstructive airway symptoms (*Parsons* and *Smith*, 1982).

The work of *Snyder* et al. (1973) on canine mandibles made it possible to apply the principle of bone elongation to the maxillofacial region which was already being widely used by *Ilizarov* in clinical orthopaedics for elongation of the long bones. However, according to *Wassmund*, in 1927 *Rosenthal* performed the first mandibular osteodistraction procedure by using an intra-oral tooth borne appliance that was gradually activated over a period of 1 month. In 1989, *McCarthy* et al., were the first to clinically apply the technique of extraoral osteodistraction on four children with congenital craniofacial anomalies. While, *Guerrero* (1990), was the first to report the results of intra-oral mandibular widening on 11 patients with transverse deficiencies ranging from 4 to 7 mm. In 1994, *McCarthy* et al., developed a miniaturized bone-borne uniguide mandibular distraction device (Howmedica Leibinger, Inc.) suitable for intra-oral placement.

Havilik and *Bartlett* (1994) and *Moore* et al. (1994) reported the treatment of severe micrognathia by using extraoral distraction devices. However, this can leave obvious facial skin scars and can easily injure the facial nerve branches. More recently, intra-oral appliances have been developed that avoid both these complications (*Howlett* et al., 1999; *Wang* et al., 2003; *Li* et al., 2008).

When compared with the extraoral distractors, the intra-oral ones have numerous advantages including: lack of external scars, less soft tissue trauma, near total concealment of the device and improved superior psychological tolerance. However, in infants and young children, there is not enough room subperiosteally to accommodate the whole distractor.

In this article, we report our experience in correction of micrognathia accompanying OSA syndrome by using internal DO techniques, utilizing a modified technique for the placement of the intra-oral distractor.

PATIENTS AND METHODS

Seven patients with mandibular micrognathia accompanying OSA syndrome were treated with bilateral mandibular DO, using an intra-oral unidirectional unburied distractor (Martin™; Germany). All patients had undergone clinical, radiographic and polysomnographic examinations preoperatively, and had been diagnosed with micrognathia (isolated Pierre Robin sequence). All patients were neurologically normal and without associated syndromes. There were 4 female and 3 male patients, with an average age of 34 months (range, 7 months–8 years). The average follow-up period was 3.7 years (range, 2–5 years) (Table 1).

Previous failed management

1. Positioning: in all patients.
2. Tongue-lip adhesion: in 2 patients.

Criteria for diagnosis of obstructive sleep apnoea (OSA)

(A) Clinically:

1. Snoring.
2. Daytime somnolence and reduced activities.

(B) Polysomnographic examination:

1. Snoring versus time.
2. Flow versus time (flattening index).
3. Less than 85% oxygen saturation.
4. Apnoea/hypopnoea index >5.

Patient examination

(A) Clinically: all patients had maxillo-mandibular discrepancy >8 mm.

(B) Radiographic analysis: the patients were assessed for surgery using panoramic views, cephalometric analysis (lateral cephalogram) and normative data for comparison of maxillo-mandibular growth and relationships. All cephalometric radiographs were traced and evaluated by a single orthodontist, and all indicated the need for 14 mm or more of distraction (range, 14–20 mm; average, 17.4) to achieve a normal maxillo-mandibular relation. However, an over-correction by about 2–3 mm was required and was the rule in all cases of this study to compensate for future retarded mandibular growth.

Postoperative radiographs were obtained for each patient following completion of the consolidation period (4 weeks) and at 3, 6, 12, and 24 months.

Follow-up panoramic radiographs were used to document new bone formation before removal of the distractor.

Objective measurements of changes in airway size (at the tongue base level) were determined from lateral cephalometric radiographs obtained pre- and post-

Table 1 — Patient review

Pt no.	Age	Sex	Pathology
1	7 mo	Male	• Severe bilateral mandibular hypoplasia. • Sleep apnea. • Difficult feeding.
2	20 mo	Female	• Severe bilateral mandibular hypoplasia. • Sleep apnoea. • Difficult feeding. • Cleft palate.
3	8 year	Female	• Severe bilateral mandibular hypoplasia. • Sleep apnoea. • Disturbed occlusion and difficult mastication.
4	20 mo	Male	• Severe bilateral mandibular hypoplasia. • Sleep apnoea. • Cleft palate. • Difficult feeding.
5	4.5 year	Male	• Severe bilateral mandibular hypoplasia. • Sleep apnoea. • Disturbed occlusion and difficult mastication.
6	5 year	Female	• Severe bilateral mandibular hypoplasia. • Sleep apnoea. • Disturbed occlusion and difficult mastication.
7	6 year	Female	• Severe bilateral mandibular hypoplasia. • Sleep apnoea. • Disturbed occlusion and difficult mastication. • Limitation of jaw movement.

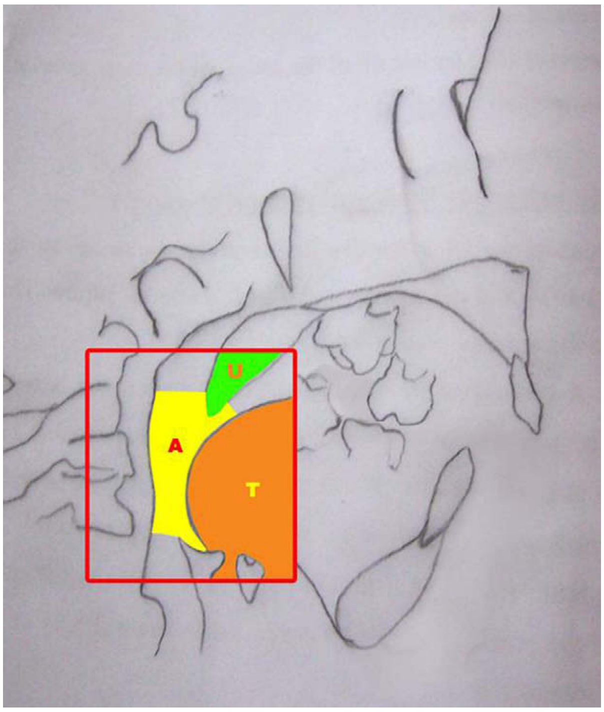

Fig. 1 — Shows the effective airway space measurement on a cephalometric tracing. A = air-way space, U = uvula, T = tongue base.

operatively according to the method utilized by *Denny et al., 2001*. The effective airway space represents a single-plane measurement defined by the following boundaries: a horizontal line from the tip of the odontoid process to the velum, the uvula tip to the tongue base along the shortest line, the tongue base down to the base of the epiglottis, and the "horizontal" line to the posterior pharynx (Fig. 1). These lines were traced for each cephalogram, and the resulting tracings were loaded into Adobe PhotoShop, version 7.0 ME (Adobe Systems-Calif, USA), using a Hewlett-Packard Jet Il-C scanner. The square area enclosed by the tracing was calculated using Image Software. The statistical analysis of changes in this square area was performed using the student's paired test.

Polysomnographic examinations (Sleep Study) of all patients were performed before operation, 1 week after activation and at the end of activation, at the Sleep and Respiratory Laboratory of Chest Department, Tanta University Hospital, Tanta, Egypt. By using of an auto set clinical system (Res Med., Australia), which consists of auto set flow generator and auto set clinical computer set, connected to a personal computer. All patients were subjected to diagnostic sleep study at night for at least 7 h. The following parameters were continuously monitored:

Snoring versus time

Snoring was displayed on a scale of 0 to 4 units. One unit of snoring is equivalent to 75 dB (decibels). A value of zero corresponds to silence; a value of 1—3 corresponds to loud snoring and a value of 4 or more indicates an extremely loud snoring. Data in this study are presented as % of time of study in which snoring index was more than 1 unit (loud snoring).

Flow versus time (flattening) index

It represents the average shape of the last five inspirations. It is used to determine upper airway resistance. The shape of the inspiratory flow-time curve in normal unoccluded breath is rounded in appearance. If there is inspiratory airflow limitation, the flow-time curve will be flattened, particularly over the middle half. An index of the curvature or flattening is derived. A typical unobstructed breath will have a curvature or flattening index more than 0.15. A severely flow limited breath will have an index of 0.1 units or less. Data in this study are presented as a % of time of study in which flattening index was less than 0.15.

Oxygen saturation

O_2 saturation is shown in a scale of 0—100%. Percentage of time of sleep with O_2 saturation less than 85% and lowest O_2 saturation were monitored.

Apnoea/hypopnoea index

Apnoea is defined as cessation of airflow for more than 10 s. A hypopnoea is scored if the average ventilation drops below approximately 50% of the recent average flow for at least 10 s. The apnoea/hypopnoea index is the number of apnoea and hypopnoea per hour of sleep study.

Statistical analysis: All results are represented in mean ± SD, one way analysis of variance is used to compare repeated different parameters in the sleep study, pairwise multiple comparisons were performed using Tukey test. $p < 0.05$ is considered significant.

The criteria for cure were as follows: (1) the disappearance of clinical symptoms (daytime somnolence and sleep snore); (2) absence of apneic attacks during sleep hours; (3) lowest oxygen saturation more than 85%, and (4) apnoea/hypopnoea index <5.

Surgical procedure and distraction protocol

The operation was performed by an intra-oral procedure. All operations were performed with the patient under general anesthesia. At the body of the mandible, a buccal corticotomy and superior and inferior osteotomies with a standard surgical reciprocating saw were performed under sterile saline irrigation.

The lingual cortex and medulla bones were disrupted by a "greenstick" maneuver with an osteotome. This procedure can effectively prevent injury to the inferior alveolar nerve. The distraction devices were fixed to the mandibles by unicortical micro screws according to the position and direction designed before operation.

The modified technique for distractor application (unburied device) allows the whole distractor to protrude through the vestibular mucosa into the buccal cavity instead of being buried subperiosteally. During the corticotomy and osteotomies, the neurovascular bundle was carefully preserved.

This technique implies that, the distractor is fixed high up on the external oblique ridge thus the plate foot is only buried subperiosteally, while the rest of the distractor remains protruding from the vestibular incision, avoiding interference with the opposing jaw (Fig. 2).

- The latency period was 3 days.
- The distraction rate was 1.0 mm with a rhythm of 0.5 mm twice a day.
- The distraction period was 17−24 days.
- The consolidation period was 4 weeks.

RESULTS

After completion of distraction, retrognathia was corrected in all 7 patients (Fig. 3). The subjective symptoms of all patients had disappeared completely or had been alleviated after completion of mandibular DO and through the follow-up period. The distraction procedure was smooth. Postoperative radiographic examination and analysis showed that good new bone formed in the distraction gap (except in 2 patients where an unequal bone formation was detected, Fig. 4), and the mean effective airway space increase was 70.5% (range, 31−105%, $p < 0.01$) when compared with the effective airway space pre-distraction (Fig. 5). Polysomnographic examination showed that after mandible distraction was completed, the apnoea/hypopnoea index was lowered from 60 (9.8−126.5) to 1.57 (0−16.4) and the sleep apnoea symptoms had basically disappeared. The mean oxygen saturation increase was from 80% to 98% post-distraction.

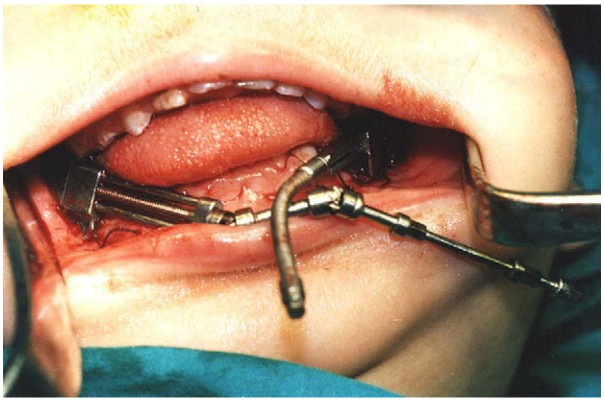

Fig. 2 − Shows intra-operative distractor placement position (a new technique).

Sleep study parameters (Table 2)

Snoring

The mean value of % of time of sleep with snoring index >1 unit before surgery, 1 week after activation and at the end of activation were 68.57 ± 7.48%, 38.57 ± 4.75% and 7.14 ± 5.66% respectively. One way repeated measures analysis of variance showed statistical significant difference ($F = 202.97$, $p < 0.001$). All pairwise multiple comparisons (Tukey test) showed that snoring index was significantly decreased 1 week after activation with further significant decrease at the end of activation compared to before surgery and 1 week after activation ($q = 13.91$, 28.49 and 14.57, respectively, $p < 0.05$).

Upper airway resistance

Flattening index less than 0.15 indicated increased upper airway resistance, the mean value of % of time of sleep with flattening index <0.15 before surgery, 1 week after activation and at the end of activation were 78.57 ± 3.78%, 49.28 ± 5.34% and 12.14 ± 6.98%, respectively. One way repeated measures analysis of variance showed statistical significant difference ($F = 399$, $p < 0.001$). All pairwise multiple comparisons (Tukey test) showed that flattening index was significantly decreased 1 week after activation with further significant decrease at the end of activation compared to before surgery and 1 week after activation ($q = 29.28$, 66.42 and 37.14, respectively, $p < 0.05$).

Oxygen saturation

The mean value of % of time of sleep with O_2 saturation <90% before surgery, 1 week after activation and at the end of activation were 37.14 ± 4.88%, 5 ± 4.08% and 49.28 ± 5.34%, 5 ± 4.08%, respectively. One way repeated measures analysis of variance showed statistical significant difference ($F = 153.69$, $p < 0.001$). All pairwise multiple comparisons (Tukey test) showed that % of time of sleep with O_2 saturation <90% was significantly decreased 1 week after activation and at the end of activation compared to before surgery ($q = 32.14$, 36.42, respectively, $p < 0.05$).

The lowest O_2 saturation before surgery, 1 week after activation and at the end of activation were 62.14 ± 3.93%,

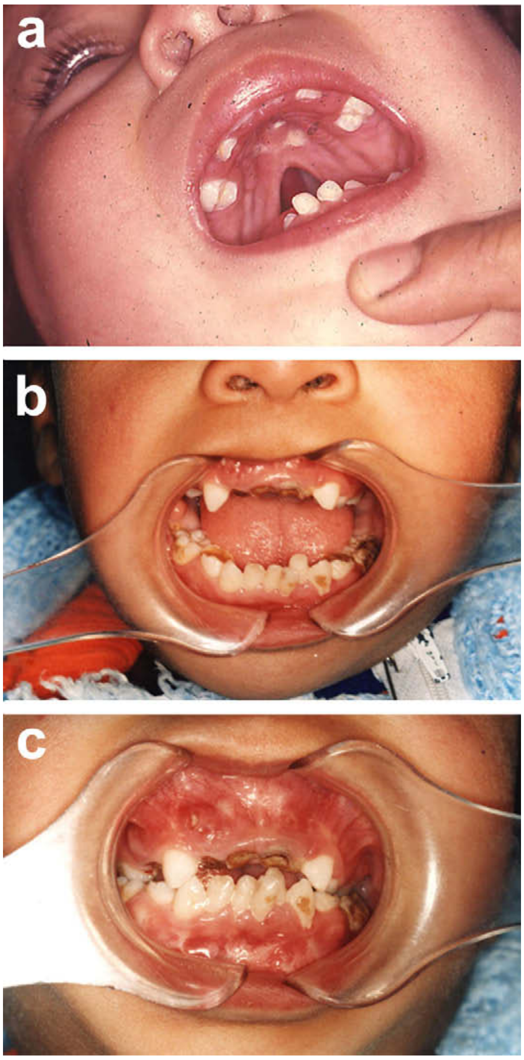

Fig. 3 – Shows the pre- and post-operative photographs of (pt no. 5) with good results achieved in long-term follow-up (4.5 years): (a) Preoperative with Pier Robin sequence. (b) Postoperative with good mouth opening. (c) Postoperative with good maxillo-mandibular relationship.

77.14 ± 4.88% and 97.57 ± 2.44%, respectively. One way repeated measures analysis of variance showed statistical significant difference ($F = 65.93$, $p < 0.001$). All pairwise multiple comparisons (Tukey test) showed that the lowest O_2 saturation was significantly increased 1 week after activation with further significant increase at the end of activation compared to before surgery and 1 week after activation ($q = 15$, 11.42 and 26.42, respectively, $p < 0.05$).

Apnoea/hypopnoea index

The mean value of apnoea/hypopnoea index before surgery, 1 week after activation and at the end of activation were 60 ± 7.25, 13.14 ± 3.02 and 1.57 ± 1.61 (events/hour) respectively. One way repeated measures analysis of variance showed statistical significant difference ($F = 313.97$, $p < 0.001$). All pairwise multiple comparisons (Tukey test) showed that apnoea/hypopnoea index was significantly decreased 1 week after activation with further significant decrease at the end of activation compared to before surgery and 1 week after activation ($q = 26.83$, 58.42 and 11.57, respectively, $p < 0.05$).

Fig. 4 – Postoperative radiograph showing an unequal bilateral mandibular bone formation at the distracted sites.

The distraction distance ranged from 17 to 24 mm, with a mean of 20.4 mm. No complications of airway obstruction or apnoea occurred post-distraction. No infections of the mandible occurred. Inferior alveolar nerve assessment is difficult in infants, but no permanent nerve injury was detected at the follow-up.

Relapse: regarding the distracted segment, during the average follow-up of 4 years (range, 2–5 years), the position of the lengthened mandible was stable and no or minimal relapse was noticed, except in one patient (pt no. 1). The onset of relapse was about 3 years later, which was accompanied by a unilateral TMJ ankylosis together with a recurrence of airway obstruction symptoms (Fig. 6). This child was reoperated upon at the age of 4.5 years with good results (Fig. 7), the first operation having been at 7 months.

Occlusion: posterior cross-bite was detected in 5 patients, whereas anterior open-bite was evident in only 2 patients (Figs. 8, 9), which was self corrected. However, after a long-term follow-up, near normal occlusion was achieved by orthodontic management (Fig. 10).

Effect on unerupted teeth: acceleration of eruption (premature eruption) was noticed in many patients, even before adequate root formation (Fig. 11), in addition to abnormal root formation, such as dilacerations, which was also reported in most of the patients (Fig. 12).

However, the most important issue was the effect of mandibular distraction on the temporomandibular joint (TMJ), which resulted in:

1. Abnormal shape of the condylar process in 3 condyles (21.4%) (Fig. 13).
2. A case of unilateral ankylosis (7.1% of the condyles) was recorded 4 years after completion of distraction (Fig. 6), which may be attributed to high forces transmitted to the growing joint during the distraction process. This child was first operated at 7 months of age, and then reoperated upon at 4.8 years of age (pt no. 1).

DISCUSSION

OSA is a serious medical problem producing both physical and behavioural derangement. It is essential to provide a thorough workup and evaluation of all patients

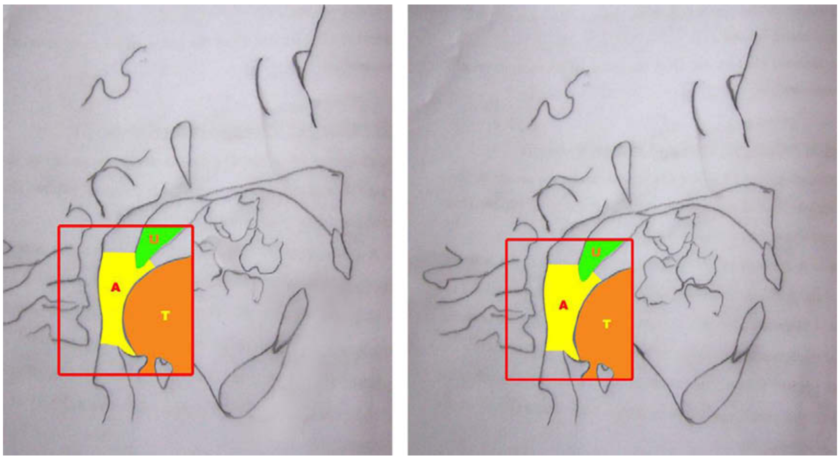

Fig. 5 — Shows the change in size of the effective airway space pre- and post-distraction.

Table 2 — Polysomnographic examination results (pre- and post-distraction)

	Pre-distraction	During distraction (1 week after activation)	Post-distraction (at the end of activation)	Statistics
Snoring versus time (mean value of % of time > 1)	68.57 ± 7.48	38.57 ± 4.75	7.14 ± 5.66	Significant
Flow versus time (flattening index) (mean value of % of time < 0.15)	78.57 ± 3.78	49.28 ± 5.34	12.14 ± 6.98	Significant
Oxygen saturation (mean value of % of time < 90%)	37.14 ± 4.88	5.0 ± 4.08	0.71 ± 1.89	Significant
Apnoea/hypopnea index (mean value)	60 ± 7.25	13.14 ± 3.02	1.57 ± 1.61	Significant

seeking care for snoring or OSA. Polysomnography is the standard for evaluation and assessment of the severity of OSA in every patient (*Aragon*, 2001).

It is now recognized that an important cause of obstructive apnoea in children is micrognathia. The size of the oropharynx is markedly reduced due to the under developed position of the mandible and corresponding retrusion of the tongue. The resulting airway obstruction may necessitate the creation of a long-term tracheostomy which is associated with a number of complications; in children, the mortality rate following tracheostomy itself (as opposed to associated condition) is around 5% (*Rogers*, 1987).

Lengthening of the mandible to correct retrognathia and the associated airway distress in patients with Pierre Robin sequence is not a new concept. The first reported surgical intervention, published by *Callister* (1937), consisted of a paediatric neurosurgical back-brace with a halo, with the infant imprisoned in this device. However, in addition to treating OSA and micrognathia successfully in 4 weeks, TMJ ankylosis followed this treatment procedure. This procedure applied traction directly to the mandible without osteotomy. As a result, the traction forces are transmitted directly to TMJ which is further immobilized by the traction appliances and accordingly this procedure was rapidly abandoned.

More recently, because of utilization of DO principles and devices which allow control of mandibular lengthening, TMJ is protected from the distraction forces. Furthermore, they allow normal joint motion throughout the treatment. TMJ ankylosis has been rare with these approaches (*Denny* et al., 2001).

Fig. 6 — Postoperative radiograph showing relapse due to a unilateral TMJ ankylosis of pt no. 1.

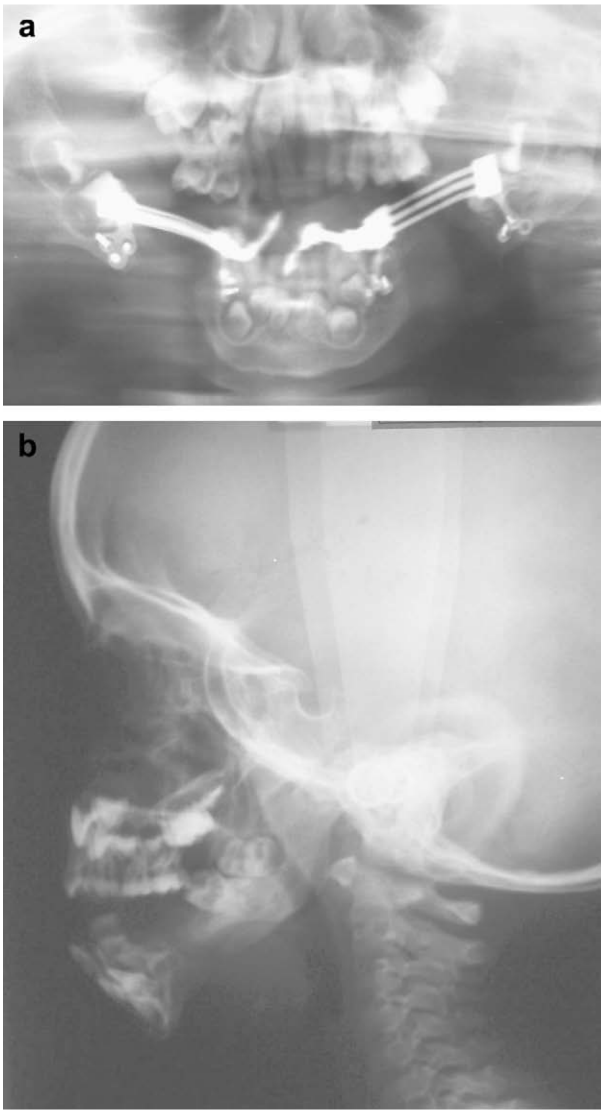

Fig. 7 – During re-distraction and post re-distraction radiograph of (pt no. 1) showing good bone formation and mandibular lengthening. (a) During re-distraction. (b) Post re-distraction.

Fig. 8 – Postoperative photograph showing posterior cross-bite.

Fig. 9 – Postoperative photograph showing anterior open-bite.

Fig. 10 – Postoperative photograph (5 years follow-up) showing near normal occlusion established by orthodontic management.

The role of mandibular distraction focuses on improving the position of the tongue in the posterior pharynx; by lengthening the mandible and bringing its muscular insertions forward, the antero-posterior dimension of the airway is thus increased (*Monasterio* et al., 2002). In this study, we successfully achieved this improvement in tongue position and the soft tissue responsible for narrowing of the posterior airway space in all patients following distraction. The improvement of airway space is evident from computerized measurements of the scanned tracings and analysis of lateral cephalograms as shown in Fig. 5. The mean effective airway space increase was 70.5% (range, 31–125%, $p < 0.01$) when compared with the effective airway space pre-distraction. This shows that soft tissue movement follows the skeletal advancement of the mandible through its muscular insertion, leading to relief and disappearance of all symptoms of OSA (as indicated clinically and by polysomnographic examinations during the follow-up period) and improvement in patients' feeding and development.

One case of relapse was recorded in this study (pt no. 1), the relapse was, in fact, due to unilateral TMJ ankylosis 3 years after completion of distraction, which was accompanied by recurrence of airway obstruction symptoms (Fig. 6). This male child was operated upon at the age of 7 months with good results and an uneventful postoperative course. He was reoperated upon successfully at the age of 4.5 years (Fig. 7). However, the exact cause of ankylosis is unknown, it may be due to high distraction forces transmitted to a joint already affected by previous

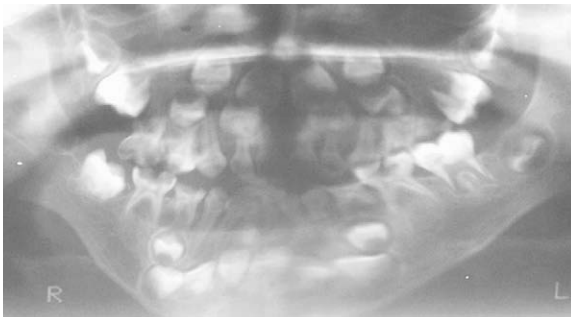

Fig. 11 – Postoperative radiograph showing premature eruption of the 1st molar at the distraction site.

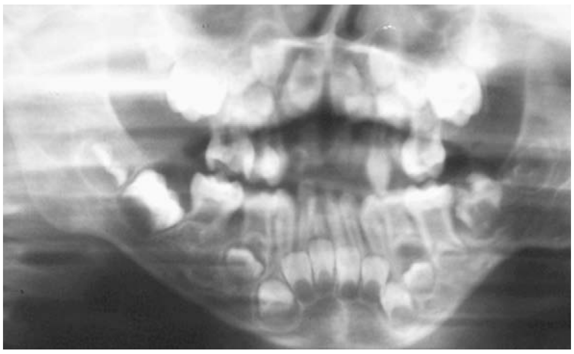

Fig. 12 – Postoperative radiograph showing abnormally formed dilacerated roots of the 1st molar at the left distracted site.

trauma or other possible factors, or more logically may be due to a blunt trauma transmitted to the joint during or after the distraction as a result of a childhood fall which passed unnoticed by his parents. Anyhow, the recurrence of (subjective) symptoms of OSA was attributed to the mandibular retrognathia as a result of retarded growth of the genetically hypoplastic mandible in comparison to normally growing maxilla which led to the development of class II maxillo-mandibular relationship.

On the other hand, an abnormal condylar shape was recorded in 3 condyles (about 21%) (Fig. 13), but without any significant effect on joint movement or mandibular growth. This could reflect the effect of distraction force transmitted to the growing condyles, which must be kept in mind during the distraction process and the follow-up periods in those young children. Therefore, we believe that encouragement of jaw movement, during and after distraction, must be stressed to the parents and child to avoid any post-distraction TMJ complications.

Acceleration of tooth eruption (premature eruption) was noticed in many patients, even before adequate root formation (Fig. 11). In addition to abnormal root formation, such as dilacerations, which was also reported in most of the patients (Fig. 12). The only damaged tooth was the lower first molar which is near to the site of osteotomy. This condition was attributed to the surgical trauma which may be responsible for stimulation of root development and tooth eruption.

The distraction distance ranged from 17 to 24 mm, with a mean of 20.4 mm. This distance was based only on cephalometric analysis in determination of degree of mandibular retrusion and maxillo-mandibular discrepancy and not correlated to the severity of OSA symptoms.

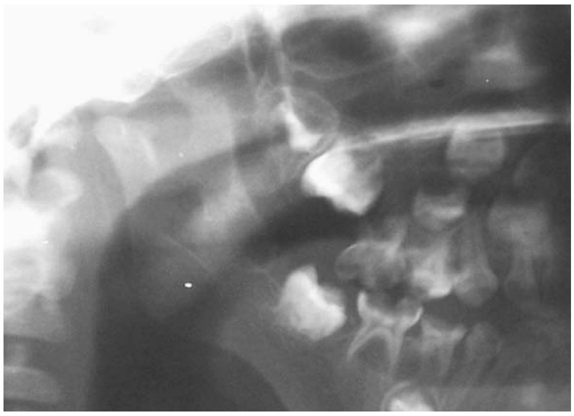

Fig. 13 – Postoperative radiograph showing abnormal shape of the Rt condylar process.

Finally, the advantages of the modified surgical technique used in this study include the following:

1. Limited periosteal stripping.
2. Less infection possibility.
3. Better monitoring of the distraction procedure.
4. Easier distractor removal.

CONCLUSION

Based on this experience, the following points were concluded:

- DO can consistently produce a worthwhile measurable and increase in the cross-sectional airway in all patients including those as young as 7 months.
- Objective measurement showed that the tongue base reliably follows the distal aspect of the mandible anteriorly.
- Mandibular distraction of 17–24 mm has eliminated the clinical symptoms of OSA in all patients.
- DO might provide an excellent alternative in cases of failure of positioning and tongue/lip adhesion in very young children.
- On long-term follow-up, DO could improve the eventual mandibular potential in those with retardation of growth.
- Re-distraction can be done safely, if needed.
- The functional airway improvement obtained is secondary to the correction of a consistent skeletal abnormality, i.e., micrognathia.
- The elimination of airway obstruction by mandibular distraction is demonstrated by clinical results and objective measurements and is independent of the syndrome causing the micrognathia with the associated airway obstruction.

References

Aragon S: Surgical management of snoring and sleep apnoea. Dent Clin North Am 4: 867–873, 2001

Bailey C, Croft C: Stertor, sleep apnoea, and velopharyngeal insufficiency. In: Scott-Brown (ed.), Otolaryngology vol (6). London: Butterworths, 350–367, 1987

Bear S, Priest J: Sleep apnoea syndrome: correction with surgical advancement of the mandible. J Oral Surg 38: 543–551, 1980

Burstein F, Cohen S, Scott P, Teague G, Montgomery G, Kattos A: Surgical therapy for severe refractory sleep apnoea in infants and children: application of the airway zone concept. Plast Reconstr Surg 96: 34−41, 1995
Callister A: Hypoplasia of the mandible (micrognathia) with cleft palate: treatment in early infancy by skeletal traction. Am J Dis Child 53: 1057−1066, 1937
Caouette-Laberge L, Bayet B, Larocque Y: The Pierre Robin sequence: review of 125 cases and evolution of treatment modalities. Plast Reconstr Surg 93: 934−942, 1994
Cohen S, Lefajyre J, Burstien F, Simms C, Kattos A, Scott P, Montgomery G, Graham L: Surgical treatment of obstructive sleep apnoea in neurologically compromised patients. Plast Reconstr Surg 99: 638−646, 1997
Cohen S, Simms S, Burstien F: Mandibular distraction osteogenesis in the management of upper airway obstruction in children. Plast Reconstr Surg 101: 312−318, 1998
Denny A, Talisman R, Hanson P, Recinos R: Mandibular distraction osteogenesis in very young patients to correct airway obstruction. Plast Reconstr Surg 108: 302−311, 2001
Guerrero C: Expansion mandibular quirurgica. Rev Venez Ortod 48(1-2), 1990
Havilik R, Bartlett S: Mandibular distraction lengthening in the severely hypoplastic mandible: a problematic case with tongue aplasia. J Craniofac Surg 5: 305−311, 1994
Hochban W, Hoch B: Obstructive sleep apnoea in the child: an interdisciplinary treatment concept with special reference to craniofacial changes. Pneumologie 52: 147−154, 1998
Howlett C, Stavropoulos M, Steinberg B: Feeding complications in a six-week-old infant secondary to distraction osteogenesis for airway obstruction: a case report. J Oral Maxillofac Surg 57: 1465−1468, 1999
Li L, Blake F, Gbara A, Gudewer E, Schmelzle R: Distraction osteogenesis in secondary microsurgical mandible reconstruction: report of seven cases. J Craniomaxillofac Surg 36: 273−278, 2008
Monasterio F, Drucker M, Molma F: Distraction osteogenesis in Pierre Robin sequence and related respiratory problems in children. J Craniofac Surg 1: 79−87, 2002
Moore M, Guzman-Stien G, Proudman T: Mandibular lengthening by distraction for airway obstruction in Treacher-Collins syndrome. J Craniofac Surg 5: 22−31, 1994
Parsons R, Smith D: Rule of thumb criteria for tongue-lip adhesion in Pierre Robin anomalad. Plast Reconstr Surg 70: 210−212, 1982
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 34: 9−18, 2005
Rogers J: Tracheostomy and decanulation. In: Scott-Brown (ed.), Otolaryngology vol (6). London: Bufferworths, 471−486, 1987
Schaefer R, Stadler J, Gosain A: To distract or not to distract: an algorithm for airway management in isolated Pierre Robin sequence. Plast Reconstr Surg 113: 1113−1125, 2004
Snyder C, Levine G, Swasson H, Browne E: Mandibular lengthening by gradual distraction: preliminary report. Plast Reconstr Surg 51: 506−508, 1973
Wang X, Wang X, Liang C, Yi B, Lin Y, Li Z: Distraction osteogenesis in correction of micrognathia accompanying obstructive sleep apnoea syndrome. Plast Reconstr Surg 112: 1549−1557, 2003
Williams J: Obstructive apnoea. In: Williams JL (ed.), Rowe and Williams Maxillofacial Injuries vol (1). London: Churchill Livingstone, 447−448, 1994

Dr Mohammad A. ELSHALL, Ph.D., Assist Prof & Consultant
Maxillofacial Surgery and Diagnostic Sciences (MDS) Department
College of Dentistry, King Saud University
Kingdom of Saudi Arabia (KSA)

Tel.: +966 56 295 8769
Fax: +966 14 679 018
E-mail: drelshall@yahoo.com

Paper received 20 October 2007
Accepted 13 October 2008