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Surgical Correction of Obstructive Sleep Apnea in Craniomaxillofacial Malformations

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ABSTRACT

Obstructive sleep apnea syndrome (OSAS) is characterized by recurrent episodes of upper airway collapse and obstruction during sleep. Patients with craniomaxillofacial anomalies usually present with OSAS secondary to decrease in volume of posterior space at the level of the hypopharynx. This study was designed to assess the improvement in OSAS by surgical correction of craniofacial malformations. The study was conducted on 14 patients presenting with different forms of craniomaxillofacial malformations and OSAS. Preoperative and postoperative evaluation of sleep apnea was done by Polysomnographic study. Surgical correction of the mandibular deficiency and/or maxillary hypoplasia greatly improved the sleep apnea symptoms.

INTRODUCTION

Obstructive sleep apnea (OSA) is characterized by recurrent episodes of cessation of breathing lasting for at least ten seconds due to upper airway collapse and obstruction during sleep **[1]**. These episodes of obstruction are associated with recurrent oxyhemoglobin desaturation and arousals from sleep. The syndrome can be a central, obstructive, or mixed type. The first form is due a decrease of impulses from the central nervous system to the diaphragm and inspiratory muscles. Obstructive sleep apnea syndrome is characterized by cessation of airflow because of upper airway obstruction despite simultaneous respiratory effort **[2]**.

The upper airway is a compliant tube and is therefore subject to collapse. The majority of patients with OSAS demonstrate upper airway obstruction, either at the level of the soft palate (nasopharynx) or at the level of the tongue (oropharynx) **[3]**. Recent research indicates that both anatomic and neuromuscular factors are important **[4]**. Anatomic factors, such as enlarged tonsils, macroglossia, or abnormal positioning of the maxilla and mandible, decrease the cross-sectional area of the upper airway and/or increase the pressure surrounding the airway, both of which predispose the airway to collapse **[5]**.

Current surgical procedures employed for OSAS include tonsillectomy and adenoidectomy **[5,6]**, nasal surgery (septoplasty, partial turbinectomy **[7]**, tongue reduction **[8]**, uvulopalatopharyngoplasty **[9,10]**. Craniofacial malformations contributing to OSAS can be treated by osteotomy advancement of the mandible and/or maxillary advancement **[11]**. Selection of surgical procedure is based on the anatomic site of the obstructive process, the severity of the sleep apnea, and the presence of skeletal abnormality.

PATIENTS AND METHODS

This study included 14 patients with different forms of craniofacial malformations. The study was conducted between May 2001 and July 2003 in the department of plastic surgery, Ain Shams University Hospitals, Cairo Egypt. The patients were in the age range of 6 months to 21-year old with an average of 11.5 years. Two patients had an Apert's syndrome, two patients had Crouzon's syndrome, four patients had maxillary hypoplasia and class III malocclusion, two patients had retrognathia and class II malocclusion, three patients had bilateral hemifacial microsomia, and one patient had bilateral temporomandibular ankylosis. Clinical assessment was done for all patients that showed variable degrees of daytime hyperexcitation, recurrent upper airway infections, restless sleep, nocturnal nightmares, and loud snoring. In the six months old patient, the only symptom was snoring or a noise emitted during nocturnal breathing.

All patients were subjected to an overnight polysomnographic evaluation in the sleep laboratory. The polysomnographic study usually includes multiple channels aiming to monitor sleep state, as well as cardiac and respiratory parameters (Fig. 1). Parameters measured during an overnight sleep study include monitoring the sleep state, electrooculogram, airflow at the nose and mouth, chest and

abdominal wall motion, pulse oximetry, electrocardiogram, and end-tidal carbon dioxide **[3,4]**.

All patients had a skull three-dimensional computed tomography that diagnosed craniofacial synostosis and the extent of midface deficiency. Cephalometry was done for all adult patients to evaluate the discrepancy between the maxilla and mandible as well as evaluation of the posterior airway space between the base of the tongue and the posterior pharyngeal wall (Fig. 2). Cephalometric radiography is however, a two dimensional method of evaluating a three-dimensional area. Thus, its reliability can be advanced by the addition of three-dimensional CT scans. Panoramic X-ray was done for patients that had hemifacial microsomia and bilateral temporomandibular ankylosis that showed a degree of bony deficiency.

Surgical procedures:

In the two patients with an Apert's syndrome, one patient underwent Fronto-orbital advancement and Le Fort III advancement osteotomy. The other patient underwent monobloc advancement (Fig. 3). In the two patients with Crouzon's syndrome, one patient underwent Fronto-orbital advancement and Le Fort III advancement osteotomy. The other patient underwent monobloc advancement.

Four patients had maxillary hypoplasia and class III malocclusion. Three patients underwent maxillary advancement. One patient underwent midface distraction. Two patients with retrognathia and class II malocclusion, one patient underwent mandibular distraction, the other patient underwent advancement genioplasty. Three patients with bilateral hemifacial microsomia underwent bilateral mandibular distraction. One patient with bilateral temporomandibular ankylosis underwent bilateral mandibular distraction with advancement genioplasty (Table 1).

RESULTS

The study included fourteen patients who had different form of craniofacial malformation and obstructive sleep apnea. Follow-up period ranged from 18 to 30 months. Postoperative cephalometry showed an increase in the posterior airway gap between the base of the tongue and the posterior pharyngeal wall. Follow-up C.T. scan showed the improvement in midface deficiency with improvement of the airway. The sleep study showed improvement in the sleep state in terms of improved airflow at the nose and mouth, better chest and abdominal wall movement, decreased end-tidal carbon dioxide, and increase oxygen saturation. Furthermore, clinical symptoms of sleep apnea improved (Figs. 3,4).

Table (1): The diagnosis, age, sex of the patients and surgical procedure performed for each patient.

Patient's diagnosis	Age (Yrs)	Sex	Surgical procedure
Apert's syndrome	2	Male	Front-orbital advancement and Le Fort III advancement osteotomy
Apert's syndrome	6 months	Male	Mono-bloc advancement
Crouzon's syndrome	21	Female	Fronto-orbital advancement
Crouzon's syndrome	5	Male	Le Fort III advancement osteotmy
Maxillary hypoplasia	12	Female	Maxillary advancement
Maxillary hypoplasia	19	Male	Maxillary advancement
Maxillary hypoplasia	13	Male	Maxillary advancement
Maxillary hypoplasia	7	Male	Midface distraction
Mandibular hypoplasia	12	Female	Mandibular distraction
Mandibular hypoplasia	15	Female	Advancement genioplasty
Hemifacial microsomia	16	Male	Bilateral mandibular distraction
Hemifacial microsomia	2	Male	Bilateral mandibular distraction
Hemifacial microsomia	17	Female	Bilateral mandibular distraction
Bilateral temporomandibular ankylosis	16	Female	Bilateral mandibular distraction with advancement genioplasty

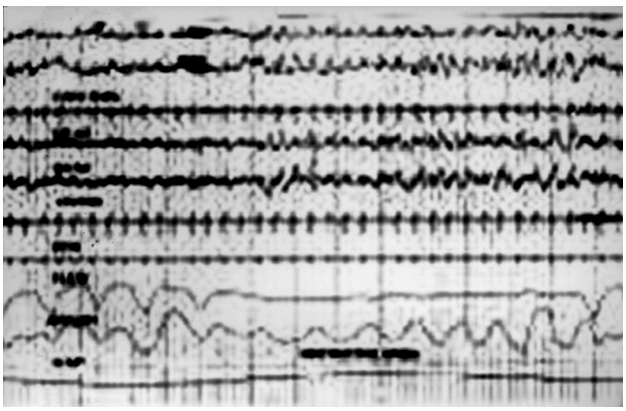

Fig. (1): Polysomnographic study showing obstructive sleep apnea. Note the absence of flow despite respiratory effort.

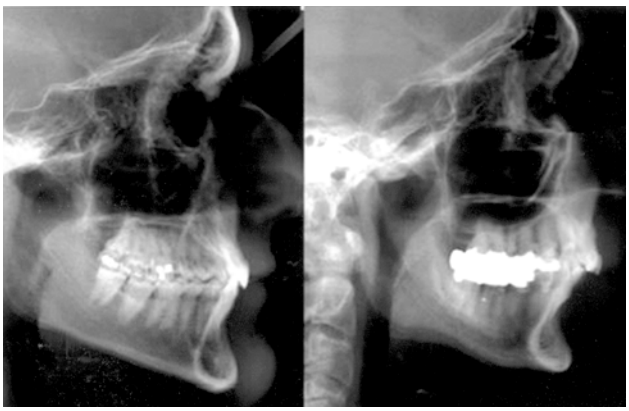

Fig. (2): (Left) Normal cephalometric radiograph. (Right) Abnormal cephalometric radiograph. Note the mandibular deficiency, elongated soft palate, and decreased posterior airway space.

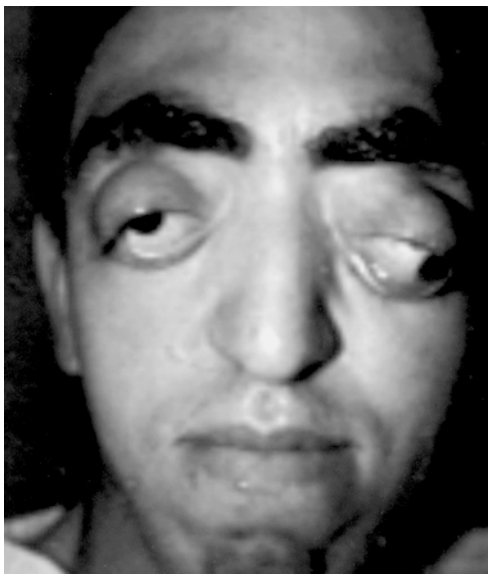

Fig. (3-A): 24-year old patient has Crouzon's syndrome.

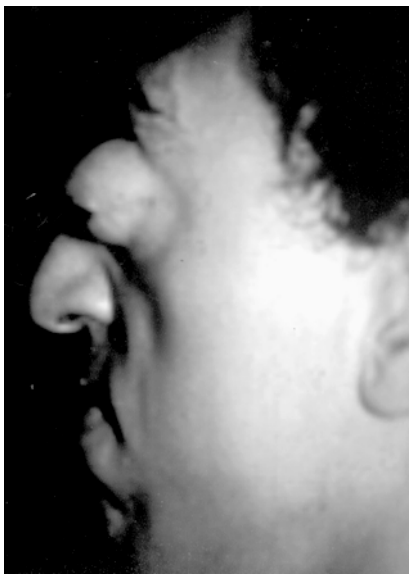

Fig. (3-B): Lateral view showing severe midface deficiency that results in sleep apnea.

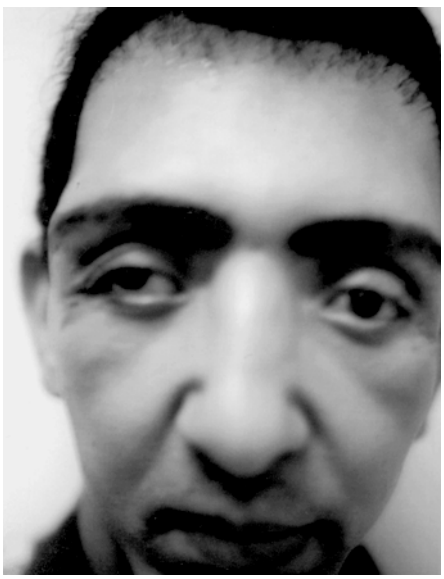

Fig. (3-C): Postoperative frontal view showing improvement in the orbital and midface areas.

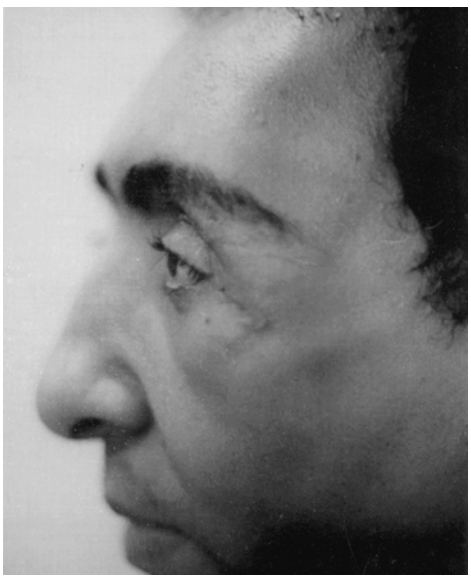

Fig. (3-D): Postoperative lateral view showing the improved midface deficiency.

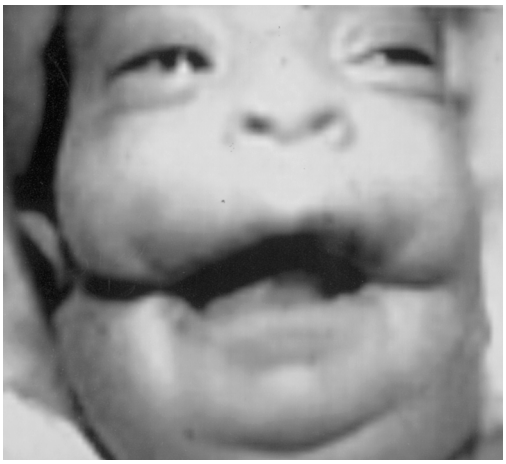

Fig. (4-A): 2-year old patient with bilateral hemifacial microsomia, preoperative front view.

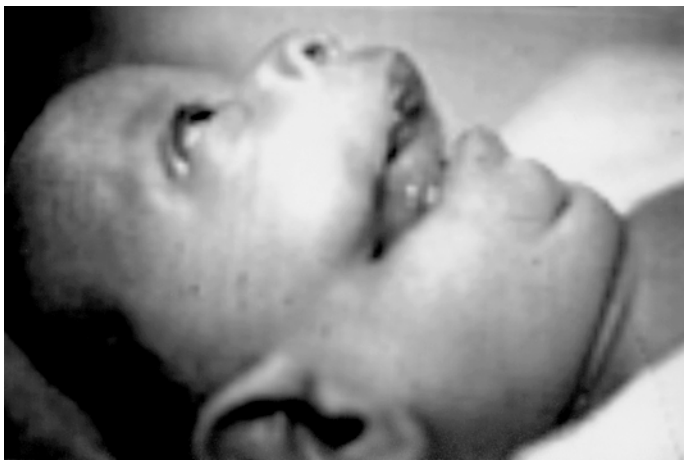

Fig. (4-B): Preoperative lateral view.

Fig. (4-C): Post closure of soft tissue and bilateral mandibular distraction.

Fig. (4-D): Postoperative lateral view.

DISCUSSION

Obstructive sleep apnea syndrome is a vital presentation in patients that have one of the craniomaxillofacial anomalies **[3]**. The potential sites of obstruction in the airway include the nose, palate (velopharynx), and tongue base (hypopharynx). Additionally, the lateral pharyngeal wall has been increasingly recognized as one of the major factors that contribute to airway collapse in obstructive sleep apnea **[12]**. With the aide of cephalometric analysis, certain craniofacial features have also been identified as risk factors in contributing to airway narrowing in OSA **[13,14]**. Today, numerous surgical procedures have been developed to improve the underlying anatomical abnormalities in treating different sites of obstruction in OSA.

Deficiency of the maxillofacial skeleton is a well-recognized risk factor of OSA **[13,14]**. Maxillomandibular advancement was initially advocated based on the finding that maxillary or mandibular deficiency or both are frequently found in patients with OSA, and that maxillomandibular deficiency results in diminished airway dimension, which leads to nocturnal obstruction. Maxillomandibular advancement achieves enlargement of the pharyngeal and hypopharyngeal airway by physically expanding the skeletal framework. In addition, the forward movement of the maxillomandibular complex improves the tension and collapsibility of the suprahyoid and velopharyngeal musculature as well as reduces lateral pharyngeal wall collapse **[15]**.

Maxillomandibular advancement is the most effective sleep apnea surgical procedure currently available. The success rate is usually between 75 and 100% with a long-term success approaching 90% **[16-21]**. Although Maxillomandibular advancement is considered a fairly invasive procedure, the associated surgical risks are low, including bleeding, infection, malocclusion, and permanent numbness. In general, patient perceptions of surgical outcome have been very favorable **[19]**.

Maxillary hypoplasia is a common finding in patients with OSA **[22,23]** and has been associated with increased nasal resistance **[24-26]**. Patients with maxillary hypoplasia often have corresponding mandibular constriction, which decreases intraoral volume. Preliminary reports on the expansion of the maxilla or the maxillomandibular complex have shown promising results in the improvement of OSA **[27,28]**. The procedure consists of limited osteotomies to allow widening of the maxilla and mandible with distractors. Maxillomandibular expansion is considerably less invasive than Maxillomandibular advancement and is easier to perform. Because orthodontic treatment is mandatory, this procedure may be best suited for adolescents and young adults.

The association between OSA and morbidity and mortality is clear. Nonsurgical treatments have been shown to be effective; however, most are limited by patient compliance. Although surgical therapy has been viewed as a treatment alternative in patients who are intolerant to conservative treatments, it is undeniable that outcome data have demonstrated that surgical therapy can be successful in the management of OSA. A thorough preoperative evaluation to identify the type of airway abnormality is mandatory. The utilization of different surgical approaches with properly selected patients can result in improved clinical outcomes while minimizing complications and avoiding unnecessary operations.

REFERENCES

1- Ancoli-Israel S., Kripke D.F. and Klauber M.R.I.: Sleep-disordered breathing in community-dwelling elderly. Sleep, 14 (6): 486-95, 1991.

2- DePonte F., Sassano P., Gennaro P., Pascali M. and Marchetti E.: Obstructive sleep apnea in a growing patient. J. Craniofac. Surg., 5: 430-434, 1999.

3- Carroll J.L. and Loughlin G.M.: Obstructive sleep apnea syndrome in infants and children: Diagnosis and management. In: Ferber R., Krgyer M., eds. Principles and Practice of Sleep Medicine in the Child. NY: McGraw Hill, 193-216, 1995.

4- Chervin R.D. and Archbold K.H.: Hyperactivity and polysomnographic findings in children evaluated for sleep-disordered breathing. Sleep, 24 (3): 313-20, 2001.

5- Sher A.E., Schechtman K.B. and Piccirillo J.F.: The efficacy of surgical modifications of the upper airway in adults with obstructive sleep apnea syndrome. Sleep, 19 (2): 156-77, 1996.

6- Friedman B.C., Hendeles-Amitai A., Kozminsky E., et al.: Adenotonsillectomy improves neurocognitive function in children with obstructive sleep apnea syndrome. Sleep, 26 (8): 999-1005, 2003.

7- Chervin R.D., Theut S., Bassetti C. and Aldrich M.S.: Compliance with nasal CPAP can be improved by simple interventions. Sleep, 20 (4): 284-9, 1997.

8- Sher A.E., Schechtman K.B. and Piccirillo J.F.: The efficacy of surgical modifications of the upper airway in adults with obstructive sleep apnea syndrome. Sleep, 19 (2): 156-77, 1996.

9- Guilleminault C.: Obstructive sleep apnea syndrome and its treatment in children: Areas of agreement and controversy. Pediatr. Pulmonol., 3 (6): 429-36, 1987.

10- Larsson H., Carsson-Nordlander B. and Svanborg E.: Four year follow-up after uvulopalatopharyngoplasty in 50 unselected patients with obstructive sleep apnea syndrome. Laryngoscope, 104: 1362-1367, 1994.

11- Riley R., Powell N.B. and Guglielminault C.: Obstructive sleep apnea syndrome: A review of 306 consectively treated surgical patients. Otolaryngol. Head Neck Surg., 108: 117-25, 1993.

12- Schwab R.J., Gupta K.B. and Gefter W.B.: Upper airway and soft tissue anatomy in normal subjects and patients with sleep-disordered breathing. Am. J. Respir. Crit. Care Med., 152: 1673-1689, 1995.

13- Jamieson A., Guilleminault C., Partinen M. and Quera-Salva M.A.: Obstructive sleep apnea patients have craniomandibular abnormalities. Sleep, 9: 469-477, 1986.

14- Partinen M., Guilleminault C., Quera-Salva M. and Jamieson A.: Obstructive sleep apnea and cephalometric roentgenograms. Chest, 93: 1199-1205, 1988.

15- Li K.K., Riley R.W., Powell N.B., et al.: Obstructive sleep apnea surgery: Patients' perspective and polysomnographic results. Otolaryngol. Head Neck Surg., 123: 572-575, 2000.

16- Li K.K., Powell N.B. and Riley R.W.: Morbidly obese patients with severe obstructive sleep apnea syndrome: Is airway reconstructive surgery a viable option? Laryngoscope, 110: 982-987, 2000.

17- Lee N.R., Givens C.D.Jr., Wilson J. and Robins R.B.: Staged surgical treatment of obstructive sleep apnea syndrome: A review of 35 patients. J. Oral Maxilofac. Surg., 57: 382-385, 1999.

18- Bettega G., Pepin J. and Veale D.: Obstructive sleep apnea syndrome: Fifty-one consecutive patients treated by maxillofacial surgery. Am. J. Respir. Crit. Care Med., 162: 641-649, 2000.

19- Li K.K., Riley R.W., Powell N.B., et al.: Obstructive sleep apnea surgery: Patients' perspective and polysomnographic results. Otolaryngol. Head Neck Surg., 123: 572-575, 2000.

20- Li K.K., Powell N.B. and Riley R.W.: Long-term results of maxillomandibular advancement surgery. Sleep Breath, 4: 137-139, 2000.

21- Conradt R., Hochban W., Brandenburg U., Heitmann J. and Peter J.H.: Long-term results after surgical treatment of obstructive sleep apnea by maxillomandibular advancement. Eur. Respir.J., 10: 123-128, 1997.

22- Seto B.H., Gotsopaulos H., Sims M.R. and Cistulli P.A.: Maxillary morphology in obstructive sleep apnea syndrome. Eur. J. Orthod., 23: 703-714, 2001.

23- Kushida C., Efron B. and Guilleminault C.: A predictive morphometric model for the obstructive sleep apnea syndrome. Ann. Intern. Med., 127: 581-587, 1997.

24- Cistulli P.A., Richards G.N., Palmisano R.G., Unger G., Berthon-Jones M. and Sullivan C.E.: Influence of maxillary constriction on nasal resistance and sleep apnea severity in patients with Marafan's syndrome. Chest, 110: 1184-1188, 1996.

25- Timms D.J.: Rapid maxillary expansion in the treatment of nocturnal enuresis. Angle Orthod., 60: 229-233, 1990.

26- Kurol J., Modin H. and Bjerkhoel A.: Orthodontic maxillary expansion and its effect on nocturnal enuresis. Angle Orthod., 68: 225-232, 1998.

27- Cistulli P.A., Palmisano R.G. and Poole M.D.: Treatment of obstructive sleep apnea syndrome by rapid maxillary expansion. Sleep, 21: 831-835, 1998.

28- Guilleminault C. and Li K.K.: Maxillomandibular expansion by distraction osteogenesis for the treatment of sleep-disordered breathing: Preliminary results. Laryngoscope, 114: 893-896, 2004.