

Variation of the upper airways in pediatric patients with OSAS and retrusion of the midface



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Variation of the upper airways to pediatric patients with OSAS and retrusion of the midface

In pediatric patients, the incidence of Sleep-Disorder breathing (SDB) is 2% for OSAS and 7-8% for snoring. Snoring, sleep apnea and the development of neurocognitive and behavioral disorders represent the main symptoms. In these children, snoring is noisy and is present for the greater part of sleep.

Accurate diagnosis and treatment protocol is critical for a child with OSAS as it is associated to complications as: pulmonary hypertension, chronic pulmonary heart disease, low height-weight development, behavioral problems, reduced school performance, bedwetting and daytime sleepiness or irritability. For this reason, over the years different surgical techniques were developed to solve the clinical symptoms evident on the polysomnographic test.

In this paper, the authors report the experience at the Department of Cranio-Maxillo-Facial Surgery, Policlinico Umberto I, "Sapienza" Università di Roma, in the treatment of pediatric patients with OSAS and midface retrusion.

KEY WORDS: Apert syndrome, Crouzon syndrome, le Fort III osteotomy and distraction, Midface retrusion, Obstruction sleep apnea syndrome, Pfeiffer syndrome

Introduction

Obstructive sleep apnea syndrome (OSAS) was first described as a nosological entity by Guilleminault in 1976¹.

Clinical features of OSAS in early childhood are irregular sleep associated with night terrors, profuse sweating, thumb sucking, sleeping in a prone fetal position (knees tucked under the belly with the buttocks up), history of recurrent respiratory infections, mouth breathing, hyperactivity, anxiety, attention deficit, snoring, repeated episodes of apnea or hypopnea associated with hypoxia and REM sleep fragmentation^{2,3}.

The prevalence of OSAS is 2% in patients between four and five years of age, and up to 40% in patients with syndromic craniofacial synostosis⁵⁻⁷.

The gold standard for the diagnosis of OSAS is polysomnography, which records two or more EEG channels, several EMG channels, movements of the chest and the abdomen, oro-nasal airflow and blood oxygen saturation.

Obstructive apnea, as mentioned previously, is a disease frequently found in pediatric patients with craniofacial malformations, particularly in children with syndromic craniofacial synostoses associated with hypoplasia of the midface, micrognathia, muscular hypotonia, cerebral compression (achondroplasia) and/or deformation of the skull base.

In these malformations, the sagittal and transverse dimensions of the facial skeleton and soft tissue atrophy lead to a reduction of the diameters of the upper airways, facilitating the collapse of these structures during sleep^{2,7}.

Surgical treatment of midface retrusion in syndromic synostoses requires a Le Fort III osteotomy with advance-

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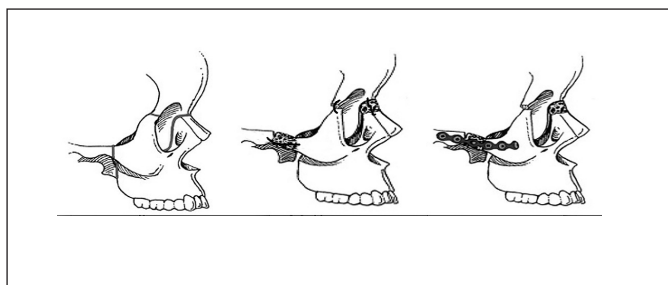


Fig. 1: The advancement of the facial skeleton by means of the Le Fort III osteotomy, obtaining an increase in sagittal and vertical dimension, getting the correction of the microorbitism and an increase in rhino-pharyngeal space.

ment of the midface, allowing for the expansion of the upper airways⁸ and also addressing issues related to the orbital district, the occlusal plane and facial aesthetics (Fig. 1).

The Le Fort III osteotomy and its advancement were first described by Gillies and Harrison⁹ and subsequently modified by Tessier¹⁰⁻¹¹. In recent years, since the first experience described by McCarthy *et al.*¹² in 1992, distraction osteogenesis has represented a paradigm shift for its application to the craniofacial skeleton and for the correction of mandibular hypoplasia. This technique was later developed and applied to midface advancement as well¹³.

The advantages of distraction osteogenesis in these patients are:

- reduced invasiveness (reduced operating time, less bleeding, no need for bone grafts for stabilization of the fracture stumps);
- reduction of relapses (the newly formed bone appears to have characteristics comparable to the those of the original);
- the simultaneous distraction of the bone and the overlying soft tissue permits multiple interventions on the facial skeleton.

In the treatment of patients with obstructive sleep apnea, the benefits of bone distraction by Le Fort III osteotomy can be ascertained from previous cases found in international literature.

While, in fact, Lauritzen *et al.*¹⁴ stated in 1986 that the Le Fort III osteotomy did not allow for the maintenance of airway patency in the postoperative period and, thus, was not a useful aid in the treatment of OSAS, in recent years, with the advent of bone distraction, several authors have instead reported satisfactory results in the treatment of these patients¹⁵⁻¹⁹.

Materials and Methods

The clinical data of twelve subjects suffering from Apert and Crouzon Syndromes were evaluated in the sample, six subjects suffering from Crouzon Syndrome and six from Apert Syndrome. Ages ranged from 5-9 years. The sample included 5 females and 7 males. From this sample, we included in our retrospective evaluation the data of those patients suffering from respiratory disorders, frequent episodes of obstructive sleep apnea, and those showing a high retrusion of the midface as assessed in cephalometric tracings.

Four patients were finally included, one with Apert, two with Crouzon and one with Pfeiffer Syndromes.

The pre-operative and post-operative 3D craniofacial CT scans, cephalograms in lateral and frontal views, and panorex films of the subjects were collected before and after surgery, during the post-operative follow-up periods of 6 months and 12 months, and at the end of the therapy. All patients underwent polysomnographic evaluation.

Cephalometric analysis of the airways and polysomnographic testing were performed before and after surgery for all patients (Tables I, II).

Results

Patient age at surgery ranged from a minimum of 28 months to a maximum of 10 years, with an average of 5.3 years.

The distraction protocol was as follows: the latency period ranged from 5 to 7 days, with an average of 5.6 days; the activation period lasted from 10 to 27 days,

TABLE I - Cephalometric Landmark used in this study

| Landmark | Name | Definition |
|----------|------------------------|---|
| S | Sella | Center of sella turcica |
| N | Nasion | Anterior point on the frontonasal suture |
| A | Point A | Deepest point on the concave outline of the upper labial alveolar process |
| PNS | Posterior Nasal Spine | The tip of the posterior nasal spine |
| ANS | Anterior Nasal Spine | The tip of the anterior nasal spine |
| U | Tip of the uvula | The most posteroinferior point of the uvula |
| UPW | Upper Pharyngeal Wall | Intersection of the line PNS-ANS and posterior pharyngeal wall. |
| MPW | Middle Pharyngeal Wall | Intersection of the perpendicular line from U with the posterior pharyngeal wall. |

TABLE II - Linear values and plans

| |
|-----------------|
| SNA |
| PNS-UPW |
| U-MPW |
| PNS-U / PNS-ANS |

with an average of 19 days; the distraction rate was 1 mm/day with two daily applications; and the consolidation phase had a duration of 5 to 6 weeks, with an average of 5.6 weeks.

Cephalometric and polysomnographic values recorded in the pre- and post-operative period are shown in Table III. The absence of polysomnographic values in patient 2 is attributable to the fact that, during this examination at another hospital, the patient underwent a tracheotomy due to cardiopulmonary arrest.

Discussion

The term “Sleep-Disordered Breathing” (SDB) includes a group of diseases ranging from simple snoring to airway obstruction²⁰. The prevalence of obstructive sleep apnea in pediatric patients varies between 1% and 3%. Additionally, it has been estimated that approximately 9% - 10% of children suffer from habitual snoring. This condition may be transient or may progress to Upper Airway Resistance Syndrome (UARS) or Obstructive Sleep Apnea Syndrome^{21,22}.

Pediatric patients with OSA have two features in particular as compared to adults:

- most of the episodes of apnea occur during REM (Rapid Eye Movement) sleep, especially in the late phase²³;
- there is a persistent obstructive hypoventilation^{24,25}.

The most common cause of obstruction of the pharyngeal region in these patients is adenotonsillar hypertrophy, due to the different growth rate of the facial skeleton with respect to lymphoid tissue^{26,27}. Other factors

that may predispose these patients to the occurrence of SDB include craniofacial anomalies²⁸, neuromuscular problems²⁹ and obesity³⁰.

In particular, in common clinical practice, there is a frequent association between SDB and craniofacial malformations, particularly in children with specific conditions such as syndromic craniofacial synostoses associated with hypoplasia of the midface, micrognathia, muscular hypotonia, and cerebral compression (achondroplasia).

In these patients the sagittal and transverse dimensions of the facial skeleton and soft tissue atrophy lead to a reduction of the diameter of the upper airways, facilitating their collapse during sleep.

In order to restore normal facial shape and treat the possible presence of SDB, over the course of several years, a series of osteotomies were designed to restore the proper sagittal and transverse diameters of the facial skeleton.

In particular, the advent and evolution of osteodistraction techniques have permitted the correction of major maxillo-mandibular defects through gradual traction of the osteotomized segments, using internal or external devices³¹⁻³² (Fig. 2). Through this method, it is possible to avoid the need for tracheotomy and facilitate decannulation of the patient^{33,34}.

The sample we reviewed consisted of patients with craniofacial malformations such as craniosynostoses (premature fusion of the growth areas of the skull), dysostoses (abnormal growth of the facial bones), or combinations of these, such as Apert, Crouzon and Pfeiffer Syndromes. Then, patients having a diagnosis of OSAS made by nocturnal cardiopulmonary recording were selected and treated by the same type of intervention, namely the Le Fort type III osteotomy with application of external bone distractors.

In the sample, persistent symptoms included snoring, sudden awakening and finally, in two patients, hypersomnia.

Treatment by Le Fort type III osteotomy provided a complete and gradual mobilization of the midface with

TABLE III - Linear values and pre/post plans

| | Patient 1 Pre / Post | Patient 2 Pre / Post | Patient 3 Pre / Post | Patient 4 Pre / Post |
|---------------------------------|-------------------------|-------------------------|-------------------------|-------------------------|
| Cephalometric values | | | | |
| SNA (°) | 75.50 / 880 | 72.50 / 800 | 62.50 / 81.50 | 760 / 890 |
| PNS-UPW (mm) | 3 mm / 17 mm | 7 mm / 16 mm | 3 mm / 17 mm | 5 mm / 16 mm |
| U-MPW (mm) | 3 mm / 7 mm | 4 mm / 6 mm | 2 mm / 8 mm | 4 mm / 7 mm |
| PNS-U / PNS-ANS (°) | 1020 / 1090 | 123.00 / 128.50 | 1080 / 117.50 | 1150 / 1220 |
| Advancement | | | | |
| Advancement of the middle third | 20 mm | 10.5 mm | 27 mm | 14 mm |
| Polysomnographic values | | | | |
| Apnea index | 30 / 6 | X / 2 | 45 / 4 | 28 / 0 |
| Apneal/hipopnea index | 55 / 11 | X / 5 | 58 / 8 | 33 / 2 |
| Sa O2 min | 78 / 90 | X / 96 | 80 / 91 | 82.5 / 97 |

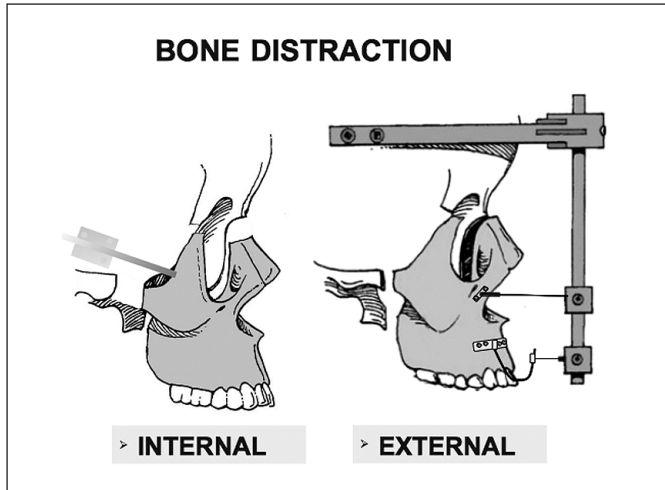


Fig. 2: In Le Fort III distraction, the distractor may be used as an external or internal device depending upon the surgical indication.

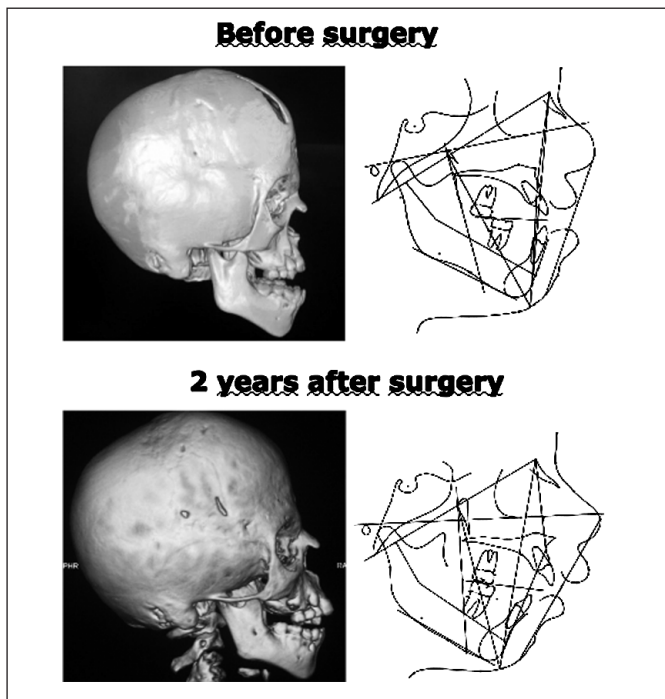


Fig. 3: 3D CT Scan and cephalometric evaluation before and after surgery in a patient with Apert syndrome, in which we used an external device after the Le Fort III osteotomy.

the simultaneous correction of the inherent problems of the airways, orbits, occlusion and facial symmetry. This osteotomy, described by Tessier, has been used in the correction of craniofacial malformations since 1967¹⁰. The same procedure is also associated with the technique of bone distraction for enlargement at the level of the nose, the palate and the pharynx and the subsequent, early removal of the tracheostomy tube³⁴ used in 17-50% of patients with craniofacial dystoses (CFD)³⁶⁻³⁷.

This treatment is usually performed in the first years of life, unless the presence of serious complications (corneal lesions, airway obstruction) do not necessitate early treatment³⁸. In the sample we studied, patient ages ranged from a minimum of 28 months to a maximum of 10 years, with an average of 3.5 years.

The same group of patients was treated with the application of a Rigid External Distractor. We generally prefer this device for its afforded ability control and vary the displacement vector during the distraction, its central distribution of the forces applied, and its easy application and removal^{20,37,40}. However, as reported in international literature, this is not very comfortable for the patient, creating both psychological and physical problems⁴¹⁻⁴³.

The timing of bone distraction used in our study provided for: a latency period of between 5 and 7 days (mean 5.6), a distraction phase lasting between 10 and 27 days (average 19 days), a rhythm of distraction of 1 mm / day with two daily applications and, finally, a consolidation phase, with a duration of between 5 and 6 weeks (average 5.6 weeks).

In our group of patients the advancement achieved with the Le Fort III, varying between 10.5 and 27 mm, allowed for a drastic reduction in episodes of apnea / hypopnea with a relative improvement of clinical symptoms and without the occurrence of major or minor complications. Moreover, the data obtained demonstrate a constant variation of the upper airways, although not directly correlated to the advancement achieved made, represented by a steady increase of the angle PNS-U / PNS-ANS and a quantitative increase in linear values represented by PNS, U-MPW and UPW.

In the treatment of pediatric patients with severe OSAS type 13, tracheotomy is reported to be an effective aid, permitting bypass of the obstructive sites. This method, as contemplated in the international literature, was applied in only one patient, who underwent a polysomnographic evaluation at another hospital and had an emergency tracheotomy due to cardiopulmonary arrest.

In our opinion, given the numerous complications possible in the peri- and post-operative periods, this type of procedure should not be considered as the first modality of treatment.

It should be noted that the necessity of using this surgical technique is mitigated through the use of therapy with continuous positive pressure ventilation (CPAP) which is well tolerated in more than 80% of patients with serious sleep-related respiratory problems, and in children with craniofacial abnormalities, obesity and neurological disorders⁴⁴.

Conclusions

Several thoughts and considerations can be drawn from the data obtained in this study. In the important disor-

ders of syndromic craniosynostosis, the severity of obstructive sleep apnea is not directly correlated with midface retrusion, therefore, a pre-operative polysomnographic evaluation is always necessary.

Given the many diseases that can present concurrently in such patients, a multidisciplinary assessment framework is essential, pre- and postoperatively.

Tracheostomy is a beneficial procedure in the treatment of pediatric patients with severe type OSAS; the same, however, should never be considered as a first-option treatment, given the possible post-surgical complications. Finally, it is important to note that the Le Fort type III osteotomy combined with distraction osteogenesis can be used in the treatment of pediatric patients with syndromic craniosynostoses such as, among others, Apert and Crouzon Syndromes and severe type OSAS, in order to reduce the severity of airway obstruction and to restore adequate symmetry.

Riassunto

L'incidenza dei disturbi respiratori del sonno, nei pazienti pediatrici, è del 2% per le OSAS e del 7-8% per i russamenti. I principali sintomi, di queste patologie, sono rappresentati dai russamenti, dalle apnee del sonno e dall'evoluzione in disordini neurocognitivi e comportamentali.

Con questo lavoro, gli Autori presentano la loro esperienza valutata nel reparto di Chirurgia Maxillo-Facciale, presso l'Ospedale Policlinico Umberto I, "Sapienza" Università di Roma, nel trattamento di pazienti pediatrici affetti da OSAS in concomitanza della retrusione del terzo medio facciale.

Sono stati presi in analisi i dati clinici di 12 pazienti affetti da Sindrome di Apert e Crouzon, di questi 6 riportavano una Sindrome di Crouzon e i rimanenti 6 una Sindrome di Apert. Il campione in esame era composto da 5 donne e 7 uomini con un'età compresa tra i 5 e i 9 anni. Gli esami radiologici sono stati effettuati prima e dopo l'intervento chirurgico, durante il follow-up eseguito a 6 e 12 mesi, e alla fine della terapia. Tutti i pazienti sono stati sottoposti a valutazione polisomnografica.

Il protocollo di distrazione è stato effettuato in un periodo di attivazione della durata di 10 - 27 giorni, con una media di 19 giorni. La distrazione veniva applicata due volte al giorno per circa 1mm al giorno. Il periodo di consolidazione è durato dalle 5 alle 6 settimane con una media di 5,6 settimane.

Le più frequenti complicanze dei disturbi respiratori del sonno sono rappresentati dall'ipertensione polmonare, dal cuore polmonare cronico, problemi comportamentali, prestazioni scolastiche scadenti, sonnolenza diurna ed irritabilità. Un'accurata diagnosi e trattamento è fondamentale nei pazienti pediatrici per prevenire le complicanze dei disturbi respiratori del sonno.

Negli anni sono state sviluppate differenti tecniche chirurgiche per risolvere questi problemi clinici.

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