Journal of Cranio-Maxillo-Facial Surgery 47 (2019) 548–555

Contents lists available at ScienceDirect

Journal of Cranio-Maxillo-Facial Surgery

journal homepage: www.jcmfs.com

Respiratory and volumetric changes of the upper airways in craniofacial synostosis patients

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ARTICLE INFO

Article history:
Paper received 16 October 2018
Accepted 28 January 2019
Available online 4 February 2019

Keywords:
Syndromic cranial-synostosis
Le Fort III osteodistraction
Frontofacial advancement
Airway volumes analysis
Polysomnography

ABSTRACT

To assess postoperative changes of the upper airway in a pediatric syndromic cranial-synostosis (SCS) population who underwent Le Fort III (LFIII) or frontofacial advancement by distraction osteogenesis (DO).

Charts' review of 25 SCS infants presented at our tertiary-care children's hospital between January 2005 and December 2016 was performed. Preoperative (T0) and postoperative (T1) three-dimensional computed tomography (3D-CT) and polysomnography (PSG) were recorded. Differences between T0 and T1 airway volumes and changes in PSG data were analyzed.

18 patients were included. The mean T0 and T1 volumes were calculated as 15.963 $mm^3 \pm$ 7.181 SD and 24.550 $mm^3 \pm$ 12.946 SD, respectively. Airway areas increased significantly ($p < 0.05$) in the total study-group by a median value of 8.004 mm^3, together with a global 72.22% improvement in respiratory parameters.

A statistically significant gain of the upper airway after LF III and DO in SCS patients has been demonstrated. Given the absence of a direct relationship between post-operative upper airway volume increase and OSAS degree improvement, further insights should consider performing T0 and T1 sleep endoscopy analysis to complete the diagnostic workup and to better assess the level of residual or recurrent upper airway obstruction in patients who experience unsuccessful surgical treatment.

1. Introduction

Craniofacial synostosis syndromes are craniofacial anomalies due to the premature fusion of the craniofacial sutures; patients affected by syndromic cranial-synostosis (SCS) (e.g.: Crouzon, Apert and Pfeiffer syndromes) commonly suffer from obstructive sleep apnea syndrome (OSAS), severe exorbitism, midfacial hypoplasia with Class III malocclusion and aesthetic issues. The cause for respiratory impairment is likely attributable to the retruded midface, which impinges on a distorted nasopharynx causing collapse of the upper respiratory tract and resulting in OSAS (Nout et al., 2012; Nixon et al., 2005) This a common feature in SCS patients, in fact, abnormal polysomnographic (PSG) values have been recently reported in literature to be present in more than 70% of these patients (Al-Saleh et al., 2011).

This pathological condition can present with different degrees of airway obstruction and it causes repeated hypopneas and apnea with snoring and hypoxia, that in some patients might lead to severe forms resulting even in failure to thrive (Boston and Rutter, 2003). Typical signs and symptoms might include daytime fatigue, malaise, irritability, sleep terrors, crying spells, growth perturbations, enuresis, delayed puberty, aggressiveness and poor eating (Nelson et al., 2008).

The management of mild to moderate respiratory impairment is usually performed by continuous positive airway pressure (CPAP), even though it is difficult to fit a mask in case of advanced forehead and maxillary retrusion to non-cooperating young patients (Fearon, 2005; Nout et al., 2008; Flores et al., 2009; Nout et al., 2010);

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https://doi.org/10.1016/j.jcms.2019.01.042

whereas, severe compromised airway obstruction might require tracheostomy and/or gastrostomy procedures. For these reasons, patients presenting severe OSAS and/or exorbitism are candidates for monobloc (MB) or Le Fort (LF) III advancement (Nout et al., 2012; Fearon, 2005; Mathijssen et al., 2006; Arnaud et al., 2007; Shetye et al., 2007). MB and LFIII osteotomy represent the traditional surgical procedures for correcting midfacial hypoplasia and they can significantly expand the pharynx and increase the upper airway volumes (Al-Saleh et al., 2011; Flores et al., 2009), even though, accordingly to the literature, about 20–30% of patients do not experience significant airway volumes increase (Saltaji et al., 2014; Xu et al., 2009; Bachmayer et al., 1986).

Whereas LFIII osteodistraction (OD) is a relatively new technique, which was first described in 1995 (Cohen et al., 1995) and then introduced as an effective and reliable technique in the management of syndromic midfacial hypoplasia, few articles have deeply analyzed and reported its short- and long-term results so far (Saltaji et al., 2014). For these reasons, the specific purpose of this study was to assess the postoperative changes of the upper airway of a pediatric population with SCS who underwent LFIII or frontofacial advancement by distraction osteogenesis to ascertain the stability of these surgical procedures. Pre-operative and post-operative three-dimensional computed tomography (3D-CT) analysis and PSG results were compared and correlated to clinical outcomes and final volumetric airway changes. Long-term results were recorded to give a whole and more complete clinical outline of our case series to better address literature discussion. Secondary aims included deriving clinically oriented insights and identifying gaps in knowledge and daily practice to stimulate future research.

2. Materials and methods

2.1. Population study

The medical records and radiological findings of all patients who underwent LF III OD or MB frontofacial advancement between January 2009 and December 2016 at Anna Meyer Children Hospital of Florence, were retrospectively identified and analyzed.

All of the patients' parents signed an informed consent agreement to allow the surgical team, comprising neurosurgeons and maxillofacial surgeons, to operate and collect children's parameters. This study was conducted in agreement with the Institutional Review Board (IRB) of the University of Florence. We followed the Helsinki Declaration guidelines.

Surgical procedures were performed by the same staff surgeons (senior maxillo-facial surgeon G.S.), under general anesthesia. Surgical indications included the presence of syndromic craniofacial malformations causing one or more of the following: airway, orbit, occlusion and facial aesthetic problems, with or without associated psychosocial problems. OSAS and severe exorbitism were the main indications for this surgery. In specific, MB advancement found surgical indication in case of severe frontal and/or cranial vault hypoplasia or synostosis with/without severe exorbitism, whilst patients who underwent Le Fort III-OD usually suffered from severe facial aesthetic issues with/without exorbitism. All of the 18 enrolled patients were affected by moderate or severe obstructive sleep apnea syndrome. On the other hand, a history of maxillofacial and/or neurosurgical procedures, presence of tracheostomy tube, the absence of pre-operative and post-operative 3D-CT images, and of PSG evaluation, incomplete clinical reports, were considered to be exclusion criteria. We also excluded patients with significant tracheomalacia, laryngomalacia, or subglottic stenosis, as those patients would still have significant airway obstruction even with craniofacial distraction osteogenesis.

Demographic and surgical/clinical data recorded were the following: age, gender, syndrome type, time of surgery, pre- and post-operative PSG results, pre- and post-operative 3D-CT findings and patient/parental observations.

Patients were seen post-operatively, 1 week after surgery, once or twice during the activation period, at the end of the activation period and at the end of the consolidation period, and then every year up to October 2018, date of our last follow-up update.

2.2. Surgical technique and distraction protocol

All the patients were operated under general anesthesia with nasal-tracheal intubation. In accordance with the protocol used by the craniofacial team, the early surgical intervention (within age 8) was required in case of severe/moderate OSAS with/without exorbitism with a risk of luxation of the eyeball; whereas children between 8 and 12 years were treated in case of exorbitism without risk of eyeball luxation or in case of a class III occlusion angle.

A standard subcranial LF III osteotomy or frontofacial advancement was performed and the midface or the frontofacial monobloc was mobilized. OD was performed with rigid external distraction device (RED; KLS Martin LP, Jacksonville, FL) that was secured to the cranium, using 3 or 4 percutaneous screws per side after the coronal incision was closed, with the plane of the device parallel to the Frankfurt horizontal. Traction wires were connected transcutaneously from the external hooks to the activating screws located on the horizontal bar of the device.

According to the literature (Fearon, 2005; Nout et al., 2008; Ishii et al., 1996), in order to achieve an adequate osteogenesis after the distraction, a latency period after the procedure was applied for a maximum of 1 week; thereafter, the device was activated at an average rate of 1 mm/day (0,5 mm in the morning and 0,5 mm in the evening) for 2–4 weeks to achieve an advancement of 15–30 mm for frontal distraction and 10–20 mm for midfacial distraction. Active distraction was followed by a fixation period of approximately twice the number of millimeters of distraction in days to allow the consolidation of the osteotomy site. Post-operative clinical evaluation was performed weekly. During the phase of distraction plain radiographs (cephalometric exams and orthopantomography) were performed to assess the progress of distraction and bone formation, and to check the position and effectiveness of the distractors. The amount of midfacial advancement was measured at the maxillary incisors on pre-operative and post-operative plain radiography.

A consolidation period of two months for the LFIII and of three months for the MB was respected.

The distractor was then removed under general anesthesia.

2.3. Imaging

For each patient, a cranio-maxillo-facial non-contrast multislice CT scan was taken at the Department of Radiology in Anna Meyer Children Hospital using the same scanner (Brilliance CT 64; Philips, Andover, MA) with a fixed slice thickness of 1.00 mm and reconstructions in the coronal and sagittal planes. General anesthesia was indicated in three cases (patient numbers 4, 10, 15) depending on patient's cooperation and age. All scans were carried out in a supine position and patients were instructed to remain still, to not swallow, to place the tongue against the incisor teeth and to hold their breath at the end of exhalation. Three-dimensional reconstructions and volumetric analysis using a computer software program (SimPlant Pro 15; Materialize Dental, Leuven, Belgium) were performed before surgery (T0), within three months after removal of the distractor (T1), and then every two years to detect the changes and stability of the upper airway. The inactive respiratory airways were manually excluded from the airway volumes analysis; the skull base identified the upper limit of the airways, while the anterior boundary was defined by a transverse plane from the uvula to the tongue base, and the lower boundary was evaluated at the tip of the epiglottis parallel to the Frankfort plane. The airway was segmented using a semiautomatic region growing method with a fixed Hounsfield threshold value. Once the 3D digital models were constructed, the airway was systematically analyzed using metric parameters (Li et al., 2003; Abramson et al., 2010; Guijarro-Martínez and Swennen, 2013; Abramson et al., 2009; Spinelli et al., 2015).

2.4. Respiratory evaluation

The respiratory evaluation was assessed using polysomnography (PSG), conducted in an accredited sleep lab and scored by a certified sleep specialist at the Meyer Children Hospital, Pulmonary Department. In patients with a tracheal cannula, preoperative PSG data were not recorded, thus they were excluded from our study.

The analysis was expressed in oxygen-desaturation-index (ODI), representing the number of desaturations (≥4% decrease with respect to the baseline) per hour; and in apnea-hypopnea-index (AHI), the number of apneas (absence of airflow for more than two breaths) and hypopneas (reduction of >50% in nasal flow signal amplitude) per hour. Accordingly to the recommendations from the American Academy of Sleep Medicine (AASM) for all indices a score of AHI over 10 was considered as a severe form of OSAS, between 10 and five as moderate, between five and one as mild OSAS, and a score less than one was considered to be normal (Kaditis et al., 2016).

A post-operative PSG study was performed at one year after the surgical procedure.

All data were entered into an SPSS database for statistical analysis (SPSS Graduate Pack, version 11.0; SPSS, Inc, Chicago, IL). Descriptive statistics including the mean and SD were calculated for the measurements obtained on 3D-CT scans. The t test for paired groups such as pre-operative (T0) and post-operative (T1) airway volumes was used to assess differences between them, and the Wilcoxon signed-rank test to document significant changes in PSG data. A p value less than 0.05 was considered statistically significant.

3. Results

Between January 2005 and December 2016 eighteen out of 25 patients fulfilled inclusion criteria for the study; in fact, of the 25 patients affected by SCS who underwent Le Fort III OD or MB advancement, two patients were excluded due to insufficient preoperative and post-operative data, and 5 patients had a preoperative tracheostomy without any pre-operative polysomnographic data.

10 out of 18 patients were males (55.5%) and 8 were females (44.5%). The median age at time of surgery was 68 months (mean 84.89 months ± 43.77 SD; range 28–195 months). The included patients were affected by Crouzon syndrome (12 patients, 66.6%) and Apert syndrome (6 patients, 33.4%). Le Fort III OD was the most frequent procedure performed (13 out of 18, 72.2%), whilst only 5 patients were treated by MB midfacial advancement (27.8%) (Table 1).

The median hospital stay was of 18 days (mean 17.61 days ± 3.68 SD; range 11–24 days). No one of the included patients had tracheostomy tube (TT) and/or naso-gastric-feeding tube (NGFT). No intraoperative nor severe post-operative complications were experienced. One local pin-site infection and one aberrant scarring were recorded over the clinical follow-up, and they all recovered well by local therapy application.

Table 1
Clinical data of the patients with respect to gender (F = female; M = male) type of cranial-synostosis syndrome, age at time of surgery and type of surgical procedure (MB = Monobloc advancement; LF III-OD = Le Fort III osteodistraction).

Patient	Gender	Cranial-Synostosis Syndrome	Age At Time Of Surgery (Months)	Surgical Procedure
1	F	APERT	67	MB
2	M	CROUZON	65	LF III-OD
3	M	CROUZON	65	MB
4	F	CROUZON	33	LF III-OD
5	M	CROUZON	112	LF III-OD
6	M	CROUZON	86	LF III-OD
7	M	CROUZON	108	LF III-OD
8	M	CROUZON	195	MB
9	F	APERT	160	MB
10	M	APERT	48	LF III-OD
11	F	CROUZON	140	LF III-OD
12	F	CROUZON	75	LF III-OD
13	F	APERT	63	LF III-OD
14	F	APERT	69	LF III-OD
15	M	CROUZON	55	LF III-OD
16	M	APERT	67	MB
17	F	CROUZON	28	LF III-OD
18	M	CROUZON	92	LF III-OD

The midfacial advancement median value registered was of 16.55 mm (mean 16.917 mm ± 3.800 SD; range, 11.2–24.1 mm); distractor device was kept between 45 and 118 days (median 62.5 days; mean 67.56 days ± 17.90 SD). All of the 18 patients received a median follow-up time of 6.5 years (mean 6.83 years ± 2.09 SD; range, 2–10 years) (Fig. 1a–c).

Pre-operative CT scans were performed on average 3 months ± 1.3 SD before the surgical procedure with a registered median volume of the starting upper airway tract of 14.77 mm^3 (mean 15.963 mm^3 ± 7.181 SD range 6.234–31.179 mm^3). Post-operative CT scans were performed on average 5 months ± 3.4 SD after the removal of the external device, reporting a median final volume value of 21.457 mm^3 (mean 24.550 mm^3 ± 12.946 SD range 6.132–54.344 mm^3), with a global median volume gain of 8.004 mm^3 (mean 23.036 mm^3 ± 66.572 SD; range from −120 to 54.344 mm^3). Post-operatively a significant pre-to post-operative increase of the airway space was found ($p = 0.04$) in 13 patients (72.22%), whereas four patients (22.22%) gained a minimal volume improvement (patients number 2, 5, 15, 18) and in one patient (5.55%) (patient number 10) a volume loss of 102 mm^3 was recorded (Table 2; Figs. 2 and 3).

Pre-operative and post-operative PSG data (ODI and AHI) were obtained on average 8.4 months ± 4.1 SD before surgery and 10.5 months ± 6.4 SD after the end of the surgical treatment, respectively. Accordingly, in the pre- and post-operative PGS data, 12 patients out of 18 (66.66%) showed an improvement of the degree of OSAS. In five patients (27.77%) the OSAS was completely resolved (patients number 3, 8, 12, 13, 16), whilst six patients (33.33%) did not experience any improvement in their OSAS degree, and two of them (33.33%) continued to be managed post-operatively with CPAP (patients number 9,10). Despite these results, all data about the PSG improvement were considered statistically significant ($p < 0.001$) (Table 3; Fig. 4).

4. Discussion

The natural progression with age of OSAS in infants has been already identified and quantified, but in patients with SCS its management requires a multidisciplinary approach, where the

Fig. 1. Pre-operative (a) and post-MB procedure (b) pictures of patient number 8. The same patient a few years later underwent rhinoplasty, mentoplasty and lipofilling additional procedures, and picture (c) represents the final result where patient is 24 years-old.

presence of central apnea does not represent absolute contraindications for surgery (Fearon, 2005; Posnick, 1997; Katz et al., 2012).

Table 2
Overview of the study population cohort regarding airway volume measurements.

Patient	Pre-operative (T0) airway volume (mm^3)	Post-operative (T1) airway volume (mm^3)	Volume gain (mm^3)
1	17.134	27.753	10.619
2	15.432	15.587	155
3	14.133	42.719	28.586
4	11.121	19.432	8.311
5	8.234	8.445	211
6	29.833	54.344	24.510
7	31.179	36.174	4.995
8	23.345	38.765	15.420
9	19.324	27.114	7.790
10	6.234	6.132	−102
11	15.123	23.342	8.219
12	11.783	17.482	5.699
13	9.321	19.572	10.251
14	16.134	27.753	11.619
15	14.432	14.582	150
16	24.122	34.752	10.630
17	12.132	19.432	7.300
18	8.334	8.534	200

Midfacial distraction osteogenesis of the craniofacial skeleton has become widely accepted and its use has increased in nasopharynx and velopharynx airway volumes since the 1990s (Nout et al., 2012; Nelson et al., 2008; Ishii et al., 1996; Meling et al., 2004; Holmes et al., 2002; Elwood et al., 2003; Meling et al., 2006; Khansa et al., 2017; Shetye et al., 2010; Wery et al., 2015). Additional surgery, such as rhinoplasty, genioplasty, and lipofilling, is usually performed after puberty depending on the growth of the craniofacial skeleton (Mathijssen et al., 2006; Gosain et al., 2002; Ponniah et al., 2008). Despite the fact that functional outcomes have shown not only upper airway obstruction improvement, but also feeding and health-related quality of life increase, there is still a lack of knowledge regarding the effects of LF III and MB advancement in SCS on the upper airway volume change and related respiratory outcomes.

Previous cephalometric studies of the upper airspace changes after large craniofacial advancements have demonstrated a significant increase in the nasopharyngeal and velopharyngeal spaces (Flores et al., 2009). In this setting, three-dimensional imaging has brought considerable advantages in clinical practice, and has become useful in planning, analyzing and monitoring surgical results (Wery et al., 2015).

On the other hand, little information is available regarding the impact of the clinical signs of OSAS and abnormal outcomes of

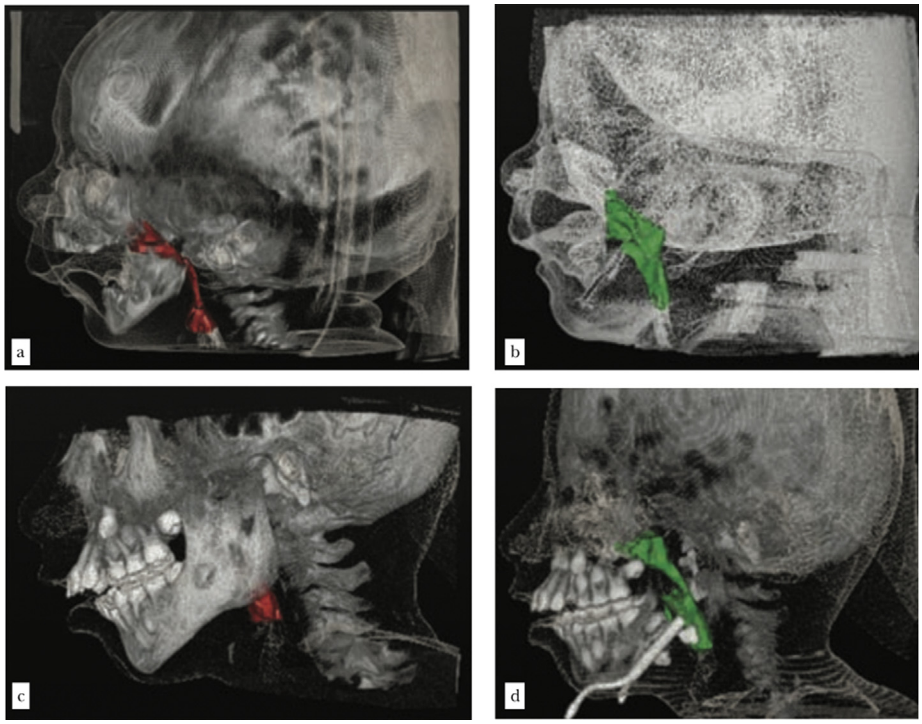

Fig. 2. Patient number 3: (a) pre-operative and (b) post-operative virtual three-dimensional computed tomography scan showing soft tissue, skull, and airway reconstruction of a young boy affected by Crouzon syndrome, who experienced a post-operative upper airway volume gain of 28.586 mm^3; patient number 6: (c) pre-operative and (d) post-operative virtual three-dimensional computed tomography scan showing soft tissue, skull, and airway reconstruction of a young boy affected by Crouzon syndrome, who experienced a post-operative upper airway volume gain of 24.510 mm^3.

polysomnography in SCS patients. It is unclear how aggressive one should be with the diagnosis of moderate/severe OSAS in order to prevent irreversible damage. It is also uncertain how much advancement is necessary to correct the OSAS (Nout et al., 2008). In addition, physicians must be aware of other structural and functional respiratory abnormalities not amenable to midface distraction such as the presence of tracheal stenosis and bronchial abnormalities, that usually coexist in syndromic patients.

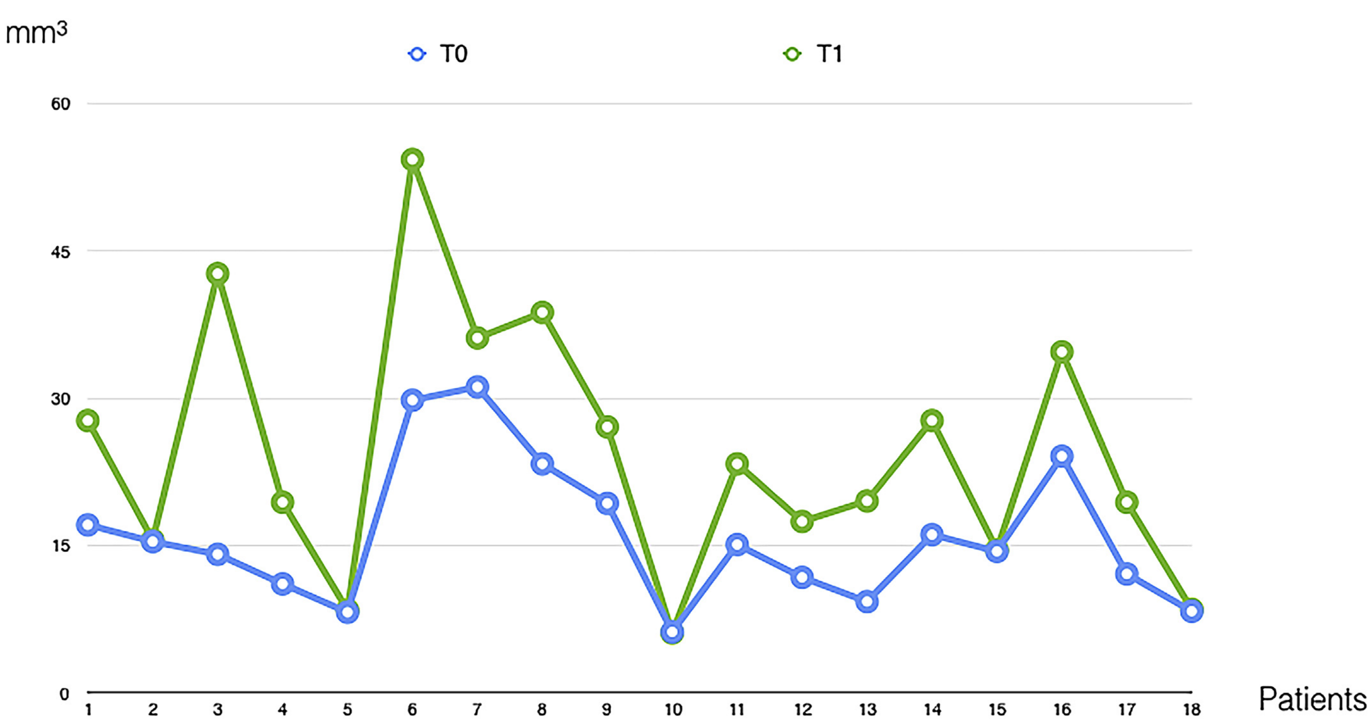

Fig. 3. Comparison between the upper respiratory airway volume expressed in mm^3 at T0 (blu) and T1 (green). The graph shows that a statistically significant increase ($p = 0.04$) in the airway volume occurred except for patients number 2, 5, 10, 15 and 18.

Table 3
Polisomnographic respiratory assessment of the study population.

Patient	Pre-op ODI	Pre-op AHI	Pre-op OSAS	Pre-op CPAP	Post-op ODI	Post-op AHI	Post-op OSAS	Post-op CPAP
1	n.r.	19	Severe	No	n.r.	4	Mild	No
2	16	24	Severe	No	15	23	Severe	No
3	3	9	Moderate	No	0	0	Absent	No
4	n.r	54	Severe	Yes	2	4	Mild	No
5	17	22	Severe	No	n.r	21	Severe	No
6	18	29	Severe	No	2	1	Mild	No
7	32	48	Severe	Yes	14	10	Moderate	No
8	n.r	7	Moderate	No	n.r	0	Absent	No
9	57	62	Severe	Yes	n.r.	20	Severe	Yes
10	34	44	Severe	Yes	33	41	Severe	Yes
11	n.r.	29	Severe	No	n.r.	7	Moderate	No
12	9	6	Moderate	Yes	0	0	Absent	No
13	4	7	Moderate	No	0	0	Absent	No
14	5	18	Severe	No	7	4	Mild	No
15	15	22	Severe	No	n.r.	17	Severe	No
16	3	7	Moderate	No	0	0	Absent	No
17	n.r.	37	Severe	Yes	2	14	Severe	No
18	15	19	Severe	No	n.r.	9	Moderate	No

(ODI = oxygen-desaturation-index; AHI = apnea-hypopnea-index; CPAP = continuous positive airway pressure; OSAS = obstructive sleep apnea syndrome; n.r. = not recorded).

Herein, through this retrospective study, we documented objective improvements in airway obstruction volumes calculated by preoperative and postoperative three-dimensional CT scans and related respiratory PSG outcomes in 18 SCS young patients who underwent cranio-facial distraction. In the present study, even though the vast majority of our patients (72.22%) improved their post-operative upper airway volume in comparison to their starting pre-operative values, a subset of 33.33% continued to have respiratory problems. By taking into consideration the complexity of the syndromic patients treated, our results reflect the current data available from the scientific literature, which declares a success rate in OSAS improvement ranging between 50% and 100% (Marcus et al., 2012; Evans et al., 2006; Lidsky et al., 2008; Meyer et al., 2008; Molina and Ortiz-Monasteiro, 1995; McCarthy et al., 1998; Cedars et al., 1999; Fearon, 2001). In this respect, the 31% of craniofacial surgeons recognize relapses of the midface after DO as the main cause of failure in their practice (Mofid et al., 2001; Kubler et al., 2004; Bannink et al., 2010). Few case series have been reported in literature analyzing LF III-OD outcomes in pediatric SCS and even fewer authors have documented upper airway volume changes through imaging measurements and respiratory outcomes comparison, so far (Nout et al., 2012; Saltaji et al., 2014; Xu et al., 2009; Meazzini et al., 2012; Meazzini et al., 2005). In our case series, despite the fact that we did not experience any relapse nor severe post-operative complications, and none of our patients required a tracheostomy procedure, the post-operative upper

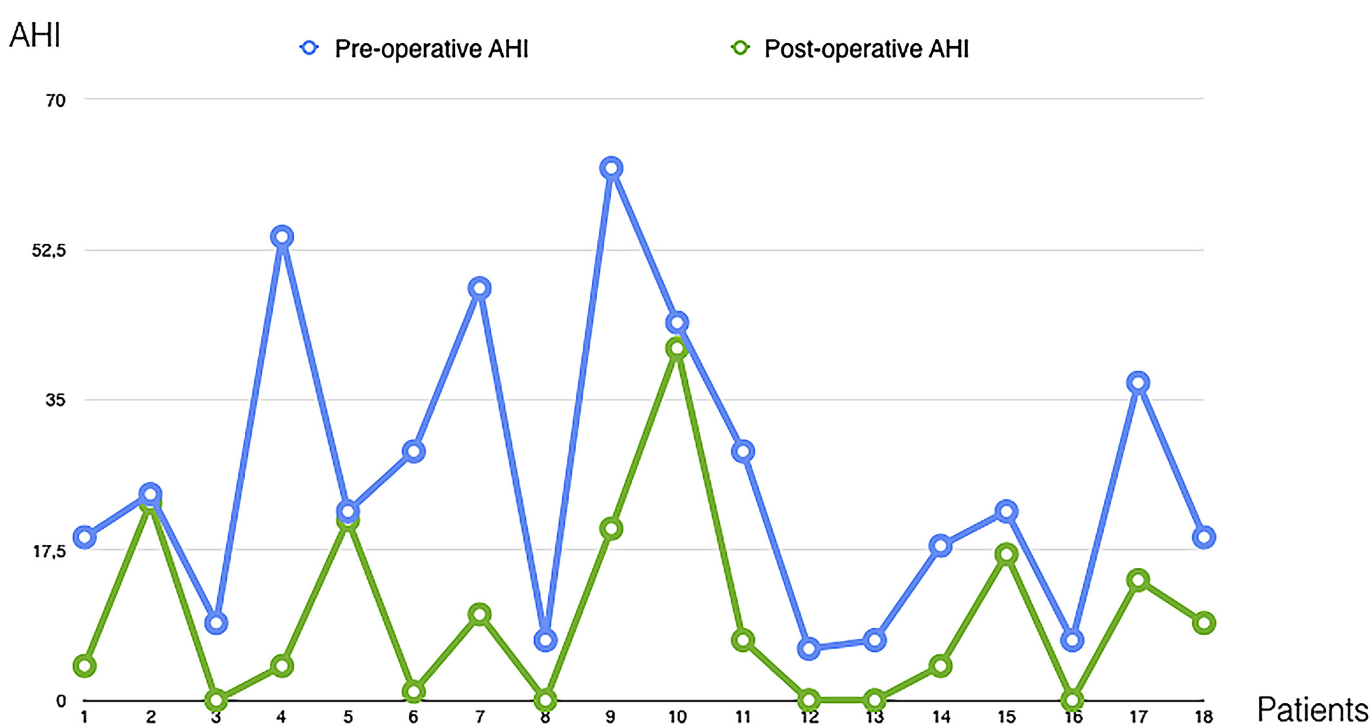

Fig. 4. Comparison between pre-operative AHI (blu) and post-operative AHI (green). The graph shows that the statistically significant improvement (p < 0.001) of these values.

airway increase was not directly related to OSAS degree of improvement. In fact, there was a global 72.22% improvement in upper airway volumes which led to a complete resolution of the OSAS in almost 27.77% of patients and to a significant betterment of obstructive respiratory symptoms in 12 cases ($p = 0.004$). Specifically, despite 9 patients (50%) experiencing a post-operative upper airway volume increase higher than the recorded global median value of 8.004 mm^3 (patients number: 1, 3, 4, 6, 8, 11, 13, 14, 16), and four other patients (22.22%) reporting an equally fair post-operative volume increase ranging between 4.995 and 7.790 mm^3 (patients number: 7, 9, 12, 17), only five of them (38.46%) resolved completely their obstructive symptoms (patients number: 3, 8, 12, 13, 16), whilst other two patients (15.38%) (patients number: 9 and 17) still remained affected by severe OSAS. On the other hand, one patient out of 18 (5.55%), although his recorded post-operative volume gain was low (200 mm^3), experienced a decrease in his OSAS degree (Table 3). It must be noted that the absence of a complete resolution of the OSAS in the whole case series can be explained by advocating its multifactorial etiology and other anatomical anomalies characterizing patients with craniofacial disorders.

Moreover, few studies in literature reported polysomnographic outcomes, and most of them did not describe time of follow-up at all (Taylor et al., 2014; Kaban et al., 1986). In addition, there is a paucity of clinical studies where polysomnographic results are evaluated and compared to pre-operative and post-operative 3D-CT findings after midfacial/frontofacial advancement as we reported herein (Nout et al., 2012; Xu et al., 2009). We first reported a longer follow-up than the most common one reported in literature, without any references to delayed failures requiring delayed tracheostomy. We did not experience any OSAS recurrence or worsening in all of the 18 patients. In addition, we applied a uniform timing of post-operative imaging study not previously published in literature. In fact, PSG data were confirmed by 3D-CT scans at T1, performed at least 3 years after the end of the surgical LF III-OD or MB treatment.

5. Conclusion

We would like to point out that there is no consensus on the growth potential of the midface after surgery, therefore, decisions and timing of surgery before skeletal maturity should be strictly bound by the indications; the absolute ones are OSAS and ocular proptosis with corneal distortion.

In this setting, 3D-CT and PSG analysis comparing the preoperative and postoperative volume and surface of the upper airway with respiratory outcomes represents a valuable tool of investigation, management and monitoring of the surgical outcome.

Because of the complexity of SCS young patients, it is inadvisable to propose any rigid surgical approach owing to their widely varying phenotype. Therefore, based on our experience, the ability to counsel SCS patients depends on our capacity to identify other sites of obstruction rather than craniofacial deformities.

Limitations of our study include the small number of patients studied and ideally, data concerning intraluminal pressure and airflow should be available. In addition, the portion of central apnea and the percentage of its improvement need to be better highlighted and addressed. Thus, we believe that a preoperative endoscopic assessment of the upper airway anatomy is recommended in order to identify any potential level of airway obstructions, associated to a neurological assessment to evaluate any potential central causes of OSAS. In case of anatomical obstruction, further orthognathic and nasal surgery (septoplasty, turbinoplasty, uvulopalatopharyngoplasty) can be indicated after midfacial and frontofacial advancement to reduce the degree of OSAS.

Funding and conflict of interest statement

None of the authors have financial conflicts or interests or otherwise to report in association with the contents of this paper.

Authorship

This manuscript was approved by all authors, and all of them have participated in the writing and correcting of this work. Further, the manuscript has not been published nor is it under consideration by other journals or editors.

Each author has participated actively in designing and writing this article, and has contributed to the following: 1) the conception or design of the work, or the acquisition, analysis, or interpretation of data; 2) drafting the work or critically revising it for important intellectual content; 3) approval of the final version to be published; and 4) agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. Specifically, Giuditta Mannelli is the main writer of the work; Giuseppe Spinelli is the main ideator of the study, performed all of the surgical procedures, and gave final approval for this version of the manuscript; Francesco Arcuri assisted in data collection and statistical analysis; Barbara Spacca helped with data collection and reviewing the final proof of the manuscript; Lorenzo Genitori provided assistance with neurosurgery surgical.

Acknowledgments

None.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.jcms.2019.01.042.

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