

CLINICAL TECHNIQUES AND TECHNOLOGY

3D computed tomographic evaluation of the upper airway space of patients undergoing mandibular distraction osteogenesis for micrognathia

Valutazione mediante tomografia assiale computerizzata 3D delle vie aeree superiori di pazienti sottoposti a distrazione osteogenetica mandibolare per micrognazia

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SUMMARY

Mandibular distraction osteogenesis (MDO) is currently an accepted method of treatment for patients requiring reconstruction of hypoplastic mandibles. To date one of the unsolved problems is how to assess the quantitative increase of mandible length needed to achieve a significant change in the volume of the posterior airway space (PAS) in children with mandibular micrognathia following distraction osteogenesis. The purpose of this study is to present quantitative volumetric evaluation of PAS in young patients having distraction osteogenesis for micrognathia using 3D-CT data sets and compare it with pre-operative situation. In this observational retrospective study, we report our experience in five consecutive patients who underwent MDO in an attempt to relieve severe upper airway obstruction. Each patient was evaluated before treatment (T0) and at the end of distraction procedure (T1) with computer tomography (CT) in axial, coronal, and sagittal planes and three-dimensional CT of the facial bones and upper airway. Using parameters to extract only data within anatomic constraints, a digital set of the edited upper airway volume was obtained. The volume determination was used for volumetric qualification of upper airway. The computed tomographic digital data were used to evaluate the upper airway volumes both pre-distraction and post-distraction. The mean length of distraction was 23 mm. Quantitative assessment of upper airway volume before and after distraction demonstrated increased volumes ranging from 84% to 3,087% with a mean of 536%. In conclusion, our study seems to show that DO can significantly increase the volume of the PAS in patients with upper airway obstruction following micrognathia, by an average of 5 times. Furthermore, the worse is the starting volume, the greater the increase in PAS to equal distraction.

KEY WORDS: 3D Computed tomographic evaluation • PAS • Mandibular distraction osteogenesis • Micrognathia

RIASSUNTO

La distrazione osteogenetica mandibolare rappresenta oggi un metodo di trattamento consolidato per i pazienti affetti da ipoplasia mandibolare. Ad oggi, un problema insoluto, nei bambini affetti da micrognazia, è la modalità di valutazione del guadagno di lunghezza mandibolare necessario ad ottenere un miglioramento significativo a livello del volume dello spazio aereo posteriore (PAS). La proposta di questo studio è la valutazione volumetrica quantitativa del PAS in giovani pazienti sottoposti a distrazione osteogenetica mandibolare per severa micrognazia, attraverso l'analisi di 'data-set' di TC pre-trattamento in comparazione ai medesimi dati post-trattamento. In questo studio retrospettivo osservazionale riportiamo la nostra esperienza relativa a cinque pazienti sottoposti a distrazione osteogenetica mandibolare. Per ciascuno dei pazienti in esame, è stata valutata la TC pre-trattamento (T0) ed al termine del trattamento (T1) nei piani assiale, coronale, sagittale e 3D a livello del PAS. Utilizzando parametri di estrazione dei dati anatomici, è stato ottenuto un modello di analisi dello spazio aereo posteriore, utilizzato per comparare le differenze volumetriche quantitative a T0 e T1. La lunghezza media di distrazione ottenuta è stata di 23 mm. L'analisi volumetrica quantitativa del PAS ha mostrato un incremento di volume, al termine del trattamento, variabile dal 84% sino 3,087% (media 536%) rispetto alla situazione pre-trattamento. Concludendo, il presente studio sembra confermare che la distrazione osteogenetica incrementi in maniera significativa il volume del PAS in pazienti con ostruzione delle vie aeree dovuta alla micrognazia. La quantificazione di tale incremento appare lineare con il guadagno ottenuto grazie alla distrazione. Nella suddetta popolazione di studio, tale guadagno è stato, in media, di 5 volte rispetto al volume di partenza. Il dato da sottolineare è che tanto minore è il volume del PAS al T0, tanto maggiore risulta il guadagno volumetrico al T1.

PAROLE CHIAVE: Tomografia computerizzata 3D • PAS • Distrazione osteogenetica mandibolare • Micrognazia

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Introduction

Upper airway obstruction occurs most commonly in individuals with craniofacial anomalies associated with

micrognathia such as Pierre Robin syndrome, hemifacial microsomia, Treacher Collins and Nager syndromes¹. In these disorders, the reduced size of mandible and its

retruded position cause retro-displacement of the tongue and concomitant reduction of the oropharyngeal airway that may lead to upper airway obstruction.

Patients have symptoms of obstructive sleep apnoea (OSA) that in severe cases require tracheal intubation and tracheostomy². More than 60% of children with craniofacial abnormalities require airway intervention as part of their overall treatment².

Tracheostomy is traditionally the safest and most effective treatment option in patients with micrognathia in Pierre Robin sequence and severe upper airway obstruction and is performed in as many as 12% of cases². Tracheostomy, however, is associated with high cost, frequent morbidity, and occasional mortality, with an average age of 3.1 years at decannulation². Therefore, the development of other effective methods of airway management is desirable.

Mandibular distraction osteogenesis (MDO) has become an accepted method of treatment for patients requiring reconstruction of hypoplastic mandibles and may achieve mandibular lengthening without need for bone graft². During the past few years mandibular reconstruction by distraction osteogenesis has been demonstrated to be effective in resolving upper airway obstruction and need for tracheostomy decannulation. Mandibular distraction has also been used in respiratory-distressed neonates and infants to avoid tracheostomy².

To date one of the unsolved problems is how to assess the quantitative increase of mandible length needed to achieve a significant change in the volume of posterior airway space (PAS) in children with mandibular micrognathia following distraction osteogenesis.

The purpose of this study is to present the quantitative volumetric evaluation of PAS in young patients having distraction osteogenesis for micrognathia using 3D-CT data sets and compared it with pre-operative situation.

Description of clinical techniques and technology

In this observational retrospective study, we report our experience in five consecutive patients (1 with Treacher Collins syndrome, 1 with Nager syndrome, 2 with bilateral hemifacial mycosomia and 1 with severe micrognathia)

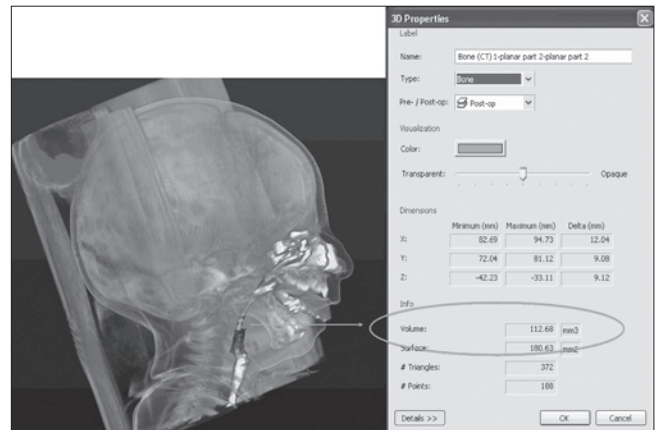


Fig. 1. Anatomic limits of upper airway in sagittal and 3D reconstructed CT scans.

who underwent mandibular DO in an attempt to relieve severe upper airway obstruction between March 2008 and May 2011 at the Maxillofacial Surgery Unit of “S. Orsola-Malpighi” University Hospital of Bologna, Italy. Inclusion criteria were the presence of syndromic or non-syndromic mandibular hypoplasia and respiratory distress with episodes of severe desaturation (oxygen saturation below 70%).

Exclusion criteria included central apnoea, apnoea that was dependent on other levels of airway impairment, such as laryngomalacia/tracheomalacia, and previous surgical procedures.

Mandibular distraction was planned bilaterally in order to advance the mandible and to increase upper airway volume. Unidirectional extra-oral (n = 4) and intra-oral (n = 1) distraction devices were used.

Each patient was evaluated before treatment (T0) and three months after the end of the distraction procedure (T1) with computed tomography (CT) in axial, coronal and sagittal planes and three-dimensional CT of the facial bones and upper airway.

Regarding CT scans, each child received the same protocol for three-dimensional CT of whole head, with 1 mm continuous axial slices, parallel to the Frankfurt horizontal³. The CT data were converted into DICOM (Digital Imaging and Communication in Medicine) format, after which the images were reconstructed for a 3D-model with

Table 1. Reference points and planes.

Area	Definition	Explanation
AREA 1 (the area between the CV ₁ and CV ₂ planes)	CV ₁	Horizontal plane passing for the most anterior point of the anterior arch of the atlas
AREA 2 (The area between the CV ₂ and CV ₃ planes)	CV ₂	Horizontal plane passing for the most anterior inferior point of the body of the 2 nd cervical vertebra
AREA 3 (The area between the CV ₃ and CV ₄ planes)	CV ₃	Horizontal plane passing for the most anterior inferior point of the body of the 3 rd cervical vertebra
	CV ₄	Horizontal plane passing for the most anterior inferior point of the body of the 4 th cervical vertebra

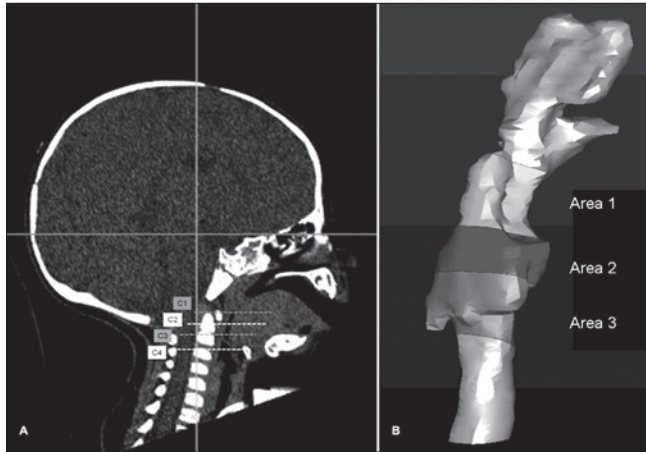


Fig. 2. 3D reconstructed CT scan volumetric assessment of the upper airway.

Simplant O&O (Materialise Dental, Leuven, Belgium). The automatically fixed threshold value was manually increased for each dataset until the nasopharyngeal airway was adequately depicted. The maximum value established as the reference threshold for this study of all patients thus defined was -306 and the minimum value was -1024.

Preparatory to upper airway analysis, the relevant reference planes and points were determined.

The upper airway was divided into three regions relative to the reference planes: area 1 (between the CV₁ and CV₂ plane); area 2 (between the CV₂ plane and CV₃ plane); area 3 (between the CV₃ and the CV₄ plane) (Fig. 1a, b; Table I) Using parameters to extract only the data within anatomic constraints, a digital set of the edited upper airway volume was obtained (Fig. 2). The volume determination was used for volumetric qualification of the upper airway. The digital CT data were used to evaluate upper airway volumes both pre-distraction and post-distraction (Fig. 3).

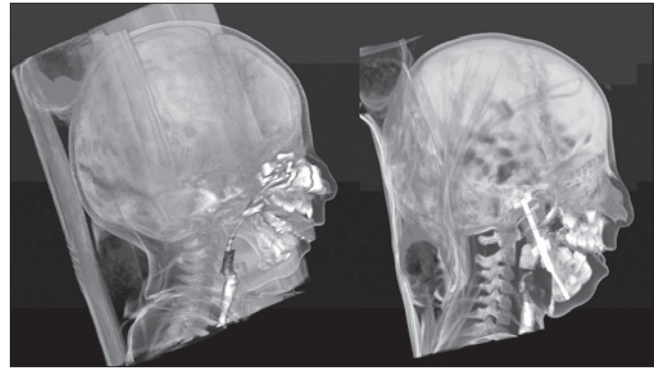


Fig. 3. 3D reconstructed CT scan showing pre-treatment airway volume (left side) and post-treatment increased volume (right side).

The present study is retrospective in nature. The study was conducted in accordance with the tenets of the WMA Declaration of Helsinki in the context of Ethical Principles for Medical Research Involving Human Subjects and was granted exemption by the local IRB of our Institution.

Results

Five infants (1 boy and 4 girls) with micrognathia, glossoptosis and severe upper airway obstruction underwent bilateral mandibular DO. The mean length of distraction was 23 mm (range, 13-35 mm) (Table II). Information on patient age, type of DO performed and the exact length of distraction for each patient is detailed in Table II.

The lateral, axial, coronal and 3D CT at the end of distraction revealed forward lengthening of the mandible and hyoid bone as exemplified in Figure 3 for all patients. These resulted in forward traction of the tongue and increased

Table II. Clinical and volumetric data.

Patient	Age	Length of DO	Vector of DO	Pre-DO upper airway volume (mm ³)	Post-DO upper airway volume (mm ³)	Δ Volume (mm ³)	Δ Volume (%)
#1	3	20 mm	oblique	A1: 113	A1: 1500	A1: +1387	A1: +1227%
				A2: 330	A2: 2194	A2: +1864	A2: +564%
				A3: 546	A3: 2017	A3: +1471	A3: +269%
#2	4	35 mm	oblique	A1: 0	A1: 3528	A1: +3528	A1: +352%
				A2: 148	A2: 4717	A2: +4569	A2: +3087%
				A3: 251	A3: 2527	A3: +2276	A3: +906%
#3	6	22 mm	oblique	A1: 811	A1: 3500	A1: +2689	A1: +331%
				A2: 635	A2: 1695	A2: +1060	A2: +166%
				A3: 351	A3: 1781	A3: +1430	A3: +407%
#4	5	13 mm	oblique	A1: 3106	A1: 7405	A1: +4299	A1: +138%
				A2: 2700	A2: 5582	A2: +2882	A2: +106%
				A3: 2670	A3: 7023	A3: +4350	A3: +162%
#5	5	25 mm	horizontal	A1: 3808	A1: 7035	A1: +3227	A1: +84%
				A2: 1485	A2: 3116	A2: +1631	A2: +109%
				A3: 2425	A3: 6098	A3: +3673	A3: +151%

Legend: A1: area 1; A2: area 2; A3: area 3.

pharyngeal space. 3D-CT demonstrated improved airway in all patients. The three-dimensional digital image demonstrated the increased airway in all areas (Table I). Area 1 volume increased between 1.387 mm³ (pt. #1) and 4.299 mm³ (pt. #3). Area 2 volume increased between 1.060 mm³ (pt. #3) and 4.569 mm³ (pt. #2). Area 3 volume increased between 1.430 mm³ (pt. #3) and 4.350 mm³ (pt. #4). The mean increased volume for all areas was 3.379 mm³ (Table II).

Quantitative assessment of the upper airway volume before and after distraction demonstrated increased volume ranging from 84% to 3,087% with a mean of 536%. All variations in terms of volumetric upper airway changes between pre and post-DO situation are detailed in Table II.

Qualitative increase of upper airway volume was assessed considering the decannulation rate at the end of the DO procedure in patients undergoing tracheostomy. In fact, all patients subjected to tracheostomy before DO underwent decannulation after the end of the described procedure. This confirms the efficacy of DO in terms of airway volume increase in patients with severe micrognathia.

During the 24-36 months of follow-up none of the patients developed symptoms of OSA and none needed CPAP treatment.

Discussion

Micrognathia is characterised by a small and retrusive mandible. Children with craniofacial anomalies associated with micrognathia, retruded position of the mandible and glossoptosis often have compromised upper airways, a condition with potential for morbidity and mortality. The degree of respiratory difficulty is dependent on the severity of the micrognathia and glossoptosis. These patients usually fail to thrive, present feeding problems, have insufficient weight gain associated with malnutrition, higher pulmonary morbidity and long-term hospitalisation¹. There are a variety of options available for airway management in the micrognathic child. Certainly, it is reasonable to begin with the most conservative measures. This includes prone positioning and placement of a nasal pharyngeal airway. The use of positive pressure mask ventilation through the nasal pharyngeal airway will provide some additional benefit. Other options include glossopexy procedures, or subperiosteal release of the floor of the mouth combined with glossopexy. For the most severe cases, tracheotomy is traditionally considered to be the definitive technique in securing a stable airway for these children². Long-standing tracheostomies are associated with high morbidity such as tracheomalacia, chronic bronchitis, laryngeal stenosis and risk of death due to the mucus plug or dislodgement of tracheostomy

tube. Patients who undergo tracheostomy require complex nursing care.

The method of DO should have a substantial advantage over all of the above mentioned techniques. It enables gradual forward advancement of the mandible and tongue that increase the pharyngeal space. The apnoea index and O₂ saturation should be markedly improved following distraction.

Most studies report improved airway and respiratory status in patients after DO based on polysomnography, cephalometry, decannulation rates and imaging studies. It is known from the literature that posterior airway volume can be accurately measured three-dimensionally using CT^{4,6}. Nevertheless, few studies have measured the extent of the increase in volume of PAS as a result of DO and no study has related the extent of distraction with the increase in volume itself. Herein, we present our quantitative assessment of increase in volume of PAS using a three-dimensional method based on CT. Our results show that the increase in volume can be much more extensive than previously assessed by other studies with similar quantities of distraction. An average increase of 536% (ranging from 84 to 3,087%) is a surprisingly positive result which gives, according to the authors, a much more important value to DO in the treatment of upper airway obstruction in mandibular micrognathia. Positive discordance with previous studies could be further investigated and can be attributed to slight differences in the evaluation method, but our analysis appears to be robust and reliable.

Furthermore, there seems to be no relation between the entity of distraction and the increase in volume. In fact, the greatest distraction (35 mm) caused the best increase in PAS (356-3087%), but the other results show no proportion with the quantity of DO. Obviously, this is only an underlying trend, because five cases are too few to evaluate a statistically significant result. Nevertheless, the slight inverse proportion between the pre-operative volume and the volume increase should be noted. This, according to our previous results in adult OSAS patients⁵, seems to demonstrate that those with greater preoperative PAS volumes can obtain lower increase in volume, independently of the entity of DO, while patients with smaller preoperative PAS volume can obtain a greater increase in volume, also independently from the entity of DO. This could mean that the possibility to augment the PAS volume varies according to the starting volume in a non-linear manner.

In conclusion, our study seems to show that DO can significantly increase the volume of the PAS in patients with upper airway obstruction following micrognathia, by an average of 5 times; the worse the starting volume, the greater the increase in PAS to equal distraction.

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