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Original Article

Skeletal changes after rapid maxillary expansion in children with obstructive sleep apnea evaluated by low-dose multi-slice computed tomography

Paola Pirelli [a,*], Ezio Fanucci [b], Aldo Giancotti [a], Michele Di Girolamo [a], Christian Guilleminault [c]

[a] Department of Clinical Sciences and Translational Medicine, University Tor Vergata, Rome, Italy
[b] Department of Diagnostic Imaging, University Tor Vergata, Rome, Italy
[c] Stanford University Sleep Medicine Division, Redwood City, CA, USA

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ABSTRACT

Objective: The objective of this study was to evaluate the skeletal effects of rapid maxillary expansion (RME) therapy performed using teeth as anchors, in obstructive sleep apnea (OSA) children, by low-dose computed tomography (CT) of the midpalatal suture opening, maxillary base width, nasal cavities width, first molar angulation and, unlike most studies in the literature, on the pterygoid processes distance.
Methods: Fourteen children (mean age 8.68 years) with OSA presenting a malocclusion characterized by upper-jaw contraction had 16-Multislice CT (MSCT) scans taken before (T0) and after (T1) RME. All exams were performed using a rigid protocol to ensure reproducibility of image collection over time, with a 16-row MSCT scanner equipped with a Dentascan reconstruction program. Scanning parameters were as follows: scout view in the anteroposterior (AP) and laterolateral (LL); 1.25-mm slice thickness with 0.6-mm collimation from the dentoalveolar and basal areas of the maxilla up to the nasal cavity, parallel to the palatal plane; 80 kV, 100 mA with an 11.25-mm table speed/rotation, rotation time 0.6 s. Matrix size was 512 $\times$ 512.
Results: Opening of the midpalatal suture was demonstrated in all cases. The results showed statistically significant T0 to T1 increments in all treated cases and clear imaging findings.
Conclusion: Use of three-dimensional (3D)-CT for follow-up studies requires a very rigid protocol to maintain reproducible positions in the scanner over time. The images confirm the real remodeling of craniofacial structure. However, to be valid such an imaging approach needs great attention to reproducibility of anatomic images over time. The changes in volume of the UA, even with a rigid protocol, cannot be affirmed with 3D-CT. There is a need to improve the definition of markers using this imaging approach when performing longitudinal studies; currently this issue is unresolved.

1. Introduction

We have reported a long-term prospective study of children with obstructive sleep apnea (OSA) with narrow maxilla treated with rapid maxillary expansion (RME) when prepubertal, and followed until adulthood, and we have shown complete elimination of the syndrome many years after treatment when reaching adulthood [1]. However, many children treated with RME do not have such a success story, and often more aggressive treatments may be needed in the peri-pubertal–pubertal period, particularly 'bone to bone expansion' which provides another opportunity to obtain enough palatal widening and maxillary upright movement to allow spontaneous counter-clockwise rotation of the mandible. As many children will not completely respond to RME performed on teeth, it is important to have good imaging of the changes obtained with the more limited approach of RME performed on teeth, which is the only valid treatment approach up until 11–12 years of age.

For many years, occlusal radiographs and lateral and anteroposterior cephalograms were the imaging approaches used to investigate and follow such children [2,3]; however, the bi-dimensional method of investigation represented a limitation as head posture may change over time and there are superimpositions of different anatomical structures leading to difficult evaluation of

* Corresponding author. Via Tomacelli 103, 00186, Rome, Italy.
E-mail address: p.pirelli@gmail.com (P. Pirelli).

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skeletal and soft tissues changes. Also, despite their usefulness, cephalometric X-rays require very systematic protocols to be helpful over time: the same equipment must be used, with X-rays being performed and read by the same operator. In recent times, multi-slice computed tomography (CT), a three-dimensional method of investigation (3D-CT), has been used to study the effects of RME treatment [4–9]. Such an imaging technique has its problems, and the risks of having non-comparative results with such a technique are known. Any change in positioning of the child between tests will lead to non-comparative images; and the difficulties encountered with cephalometrics with respect to reproducibility of image over time may be even more difficult with 3D-CT than with cephalometric X-rays. However, if a very rigid protocol is used each time that images are obtained, always using the same operator and paying great attention to the head positioning and always using the same guidelines, 3D-CT may bring valid information when following children treated with RME based on teeth.

The aim of our study was to evaluate the effects of RME with teeth anchorage using low-dose CT scans to study the skeletal effects of the maneuver, which led to functional improvement in children with OSA [10,11]. Following a very rigid protocol during the imaging sessions, which implies use of the same equipment over time, keeping track of head and body positions for each imaging session, giving exactly the same directions to the child, with each session involving the same experienced operator, repeat images were obtained over time. The following parameters were more specifically selected when investigating the results of the expansion: maxillary suture width at anterior, middle and posterior level; nasal width; right and left molar angulation; and the pterygoid processes distance – a parameter rarely looked at, but significant in the demonstration of the skeletal effect of the maneuver. This study was approved by the Ethical Committee of 'Tor Vergata' University (Rome, Italy), and informed consent was obtained from the parents or guardians of all patients.

2. Materials and methods

2.1. Subjects

Children presenting with snoring and clinical symptoms evoking abnormal breathing during sleep, upper jaw contraction at clinical evaluation amenable to RME, diagnostic polysomnography (PSG) performed and scored following the 2007 American Academy of Sleep Medicine (AASM) recommendations, demonstrating the presence of OSA, were considered for the study.

Patients with body mass index (BMI) > 24 kg/m^2 (overweight/obese children), syndromic children, enlarged adenotonsils, and children younger than nine years of age (to eliminate any controversy concerning the probabilistic effects of ionizing radiation, i.e. the stochastic radiation effect), and children with associated medical, neurological or psychiatric problems were excluded for consideration in the study.

For inclusion in this study, OSA participants must have undergone a pre- and post-RME PSG, and must have had a pre- and post-RME imaging study not only of good quality, but also with very strict adherence to the protocol of positioning of the child. The selected sample for the study consisted of 14 patients in mixed dentition with a mean age of 10.5 years (range 9–12 years), with a mean apnea/hypopnea index (AHI0 of 14.1 (±2.4)), and a mean lowest oxygen saturation of 75.8 (±8.3)%.

Subjects were submitted to CT evaluation before beginning RME (T0), and after the end of active expansion (T1) with the device still placed in the mouth, we made specific evaluations of the following parameters: maxillary suture width at anterior, middle and posterior level; nasal width; right and left molar angulation; pterygoid processes distance.

2.2. Data collection

To obtain reliable data over time, as mentioned above, great effort was made to follow a rigid protocol to place each child in the same position, including tongue position in the mouth, and at the same phase of the respiratory cycle for each study. Overall, each patient lay on the table with the Camper's plane perpendicular to the ground. The perpendicular light beams of the machine were used to standardize the head position in three planes: the longitudinal light beam passing through the center of glabella, and the transverse light beam passing through the lateral eye canthus. Axial CT scans were made parallel to the palatal plane, including the dento-alveolar area and the maxillary base, up to the lower third of the nasal cavity.

The scanning technique consisted of a preliminary scan performed in the anteroposterior (AP) and laterolateral (LL) projections, with the following acquisition parameters: 80 kV, 10 mAs [12]. Each patient underwent same CT examination at T0 and T1.

The CT scans were taken with a 1.25-mm slice thickness, 0.6-mm interval, 11.25-mm table speed/rotation, 80 kV, 100 mAs, 13.7 cm field of view (FOV), 512 × 512 matrix and 0° gantry angle [13,14].

All acquired data were transferred to an Advantage Windows workstation (GE Advantage) equipped with software for multiplanar and dentascan recostructions. Two-dimensional reformatted images were generated and measured by the computerized method.

Numerical evaluation of the various parameters was based on the identification and registration of a number of reference points identified on the CT images reformatted on different planes.

The material was measured twice, by the same author with at least a 1-week interval between T0 and T1.

The following parameters were measured in millimeters:

(1) Suture opening was measured at three levels on the axial plane: anterior edge, middle and posterior nasal spine.
(2) Maxillary base width was calculated on the axial plane between the vestibular border of buccal cortical plate (left and right respectively). The points were joined using a line tangent to the dental root of the first molar.
(3) Nasal width was measured on the coronal plane: the points of lateral limits were joined using a line parallel to the baseline in order to determine the maximum width of the nasal cavity.
(4) Right and left molar angulation were measured on the coronal plane, between a line passing through the cusp tips of the mesio-buccal cusp on the maxillary first molar and the apices of the palatal roots of the first molar (left and right respectively), and a line parallel to the sagittal line.
(5) The distance between the apices of the pterygoid processes (left and right) was calculated on the axial plane.
(6) At T0 the measurement of the midpalatal suture at three different levels was considered equal to 0 in order to level the different values before RME treatment that was in range of 0–0.3.

The normality of the data was evaluated using the Shapiro–Wilk test. Measurements for each patient before and after treatment were compared with Wilcoxon's paired matched test. Values are expressed as mean ± standard deviation. As a threshold of statistical significance, a value of $p \leq 0.05$ was used.

3. Results

The demographics and PSG data of the 14 children studied here are presented in Table 1.

In the 14 cases, we obtained an opening of the midpalatal suture with the resulting effects at different levels. Table 2 shows all the results in each patient of the sample.

3.1. Midpalatal suture

In all cases we obtained the opening of the midpalatal suture (Fig. 1). The increase at the anterior level of the suture showed an average opening of 4.1 mm. This increase is evident with the appearance of an interincisive space, the hallmark of the midpalatal suture opening that was always present in all cases; 3.1 mm at the medium level of the suture; 1.95 at the posterior level of the suture.

3.2. Maxillary width

RME therapy is responsible for the expansion of the maxilla with an average cross-sectional increase of 3.5 mm (Fig. 2). There were individual variations although all values showed clear differences between T0 and T1, indicating that, in all patients, the maneuver had an expansive effect.

Table 1
Demographics and polysomnography data of the patients.

	T0	T1
AHI	14.1 ± 2.4	0.5 ± 1.3
Nadir SpO_2 (%)	75.8 ± 8.3%	96.1 ± 1.8%

Fourteen patients (five male, nine female), mean age 8.6 years, mean body mass index = 22.3 ± 1.2 kg/m^2. AHI, apnea–hypopnea index; SpO_2, peripheral capillary oxygen saturation.

3.3. Nasal cavities

In all treated cases we obtained the increase in the nasal cavity width. The increase in maxillary cross-section is also shown by the study of the nasal cavities which are widened by the manoeuvre, with an average increase of the pyriform opening of 2.43 mm.

3.4. First molar angulation

Expansion of the skeletal structures was associated with a minimal tipping of the maxillary first molar or second deciduous molar. Tipping for the teeth occurred in all subjects but was not identical in the right and the left sides; the changes for the right molar being an average 3.6°, and for the left molar an average of 3.7°.

3.5. Pterygoid processes

From the study of the pterygoid processes distance, we found an average increase of 2.6 mm (Fig. 3).

In summary, the midpalatal suture was opened in all patients. A statistically significant difference was observed between the measurements of the maxillary, nasal cavities, and pterygoid processes distance width, performed before and after treatment. The mean amplitude of the maxilla in T0 was 51.6 ± 2 and in T1 was evaluated at 55.1 ± 3, $p = 0.0002$. The nasal cavities measurement was 29.07 ± 2 pre-treatment, and 31.5 ± 2 post-treatment, $p = 0.0001$. The mean amplitude of the pterygoid processes distance was 51.7 ± 3 pre-treatment, and 54.03 ± 3 post-treatment, $p = 0.0001$.

These CT changes were associated with changes in PSG findings with an AHI = 0.5 (±1.3) and lowest saturation = 96.1 (±1.8) %.

Table 2
Results in each patient of the sample.

Patient	Anterior suture	Middle suture	Posterior suture	Nasal cavities	Pterygoideus processes	Maxillary width	Right angle	Left angle
Before RME								
1	0	0	0	30.6	50.1	52.7	36.4	35.5
2	0	0	0	23.6	44.2	47.2	28.2°	28.2°
3	0	0	0	31.5	60.8	52.4	41.3°	32.7°
4	0	0	0	27.2	48.4	48.1	39.5°	27.2°
5	0	0	0	31.2	49	54.9	30.7°	32.6°
6	0	0	0	30.2	50.7	51.7	39.1°	36.7°
7	0	0	0	26.9	52.7	53.7	33.5°	30.1°
8	0	0	0	31.5	52.6	52.7	29.7°	33.7°
9	0	0	0	26.9	49.7	57.4	25.4°	30.5°
10	0	0	0	31.2	55.7	53.6	44.4°	38.4°
11	0	0	0	32	51.2	49.6	40°	36.7°
12	0	0	0	28.7	54.6	47.7	36.1°	36.1°
13	0	0	0	31.5	54.9	50.1	30.1°	28.4°
14	0	0	0	24.1	50.4	51.8	29.3°	32.4°
After RME								
1	2.7	2.4	1.4	31.9	50.8	54.1	40.3°	41.1°
2	6.2	5.6	5.1	29.9	48.4	53.4	31.2°	31.5°
3	4.1	3	1.8	33.8	61.8	56.7	48.1°	33.7°
4	2.7	2	1.7	28.4	52.3	51.2	39.4°	35°
5	2.7	2.2	0.8	33.1	52.6	56.3	35.8°	39.6°
6	3.4	3.3	1.4	32.9	51.9	54.3	43.6°	40°
7	5.7	4.2	2	34.2	54.1	58.3	33.6°	36.3°
8	4.9	3.5	2.9	34.1	54.2	57.7	38.9°	35.9°
9	3.7	3	1.5	28.7	50.7	60.8	31.3°	34°
10	2.3	1.8	1	31.9	56.7	53.6	48°	36°
11	3.2	2.8	2	32.4	52.5	50.7	41.9°	37.6°
12	5.8	2.8	1.2	29.9	59.2	50.4	38.1°	37.9°
13	5.8	4.4	3	33.9	60.1	55.7	32.3°	32.4°
14	5.4	4.5	1.5	27.2	55.9	58.2	44.9°	39.5°
Overall mean value								
T0	0	0	0	29.7	51.7	51.6	35.5°	32.8°
T1	4.1	3.1	1.95	31.5	54.3	55.1	39.1°	36.5°
Difference	4.1	3.1	1.95	2.43	2.6	3.5	3.6°	3.7°

RME, rapid maxillary expansion.

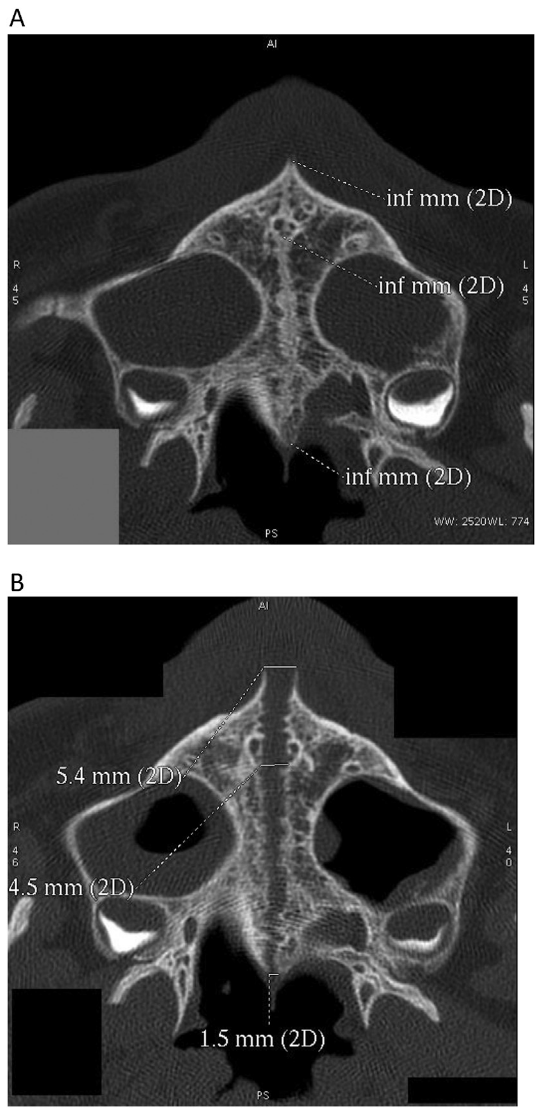
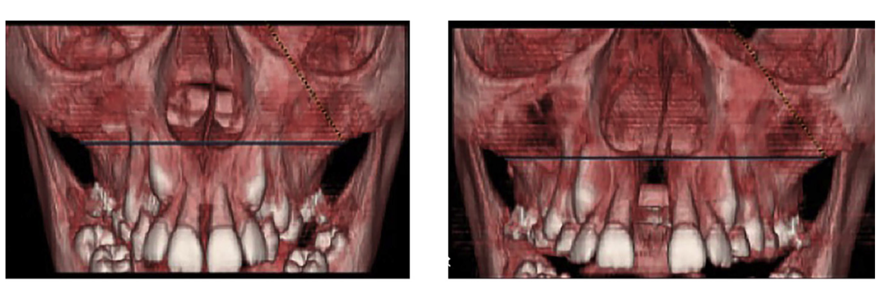


Fig. 1. A Midpalatal suture before RME. B Midpalatal suture opening after RME, measured at 3 levels: anterior, medium, posterior.

4. Discussion

RME anchored on teeth is performed more and more in young children with OSA as the presence of an abnormal narrow palate is frequently noted with or without enlarged adenotonsils [15,16], particularly after the demonstration of incomplete results of tonsillectomy and adenoidectomy (T&A) surgery and reoccurrence of abnormal breathing during sleep post T&A [17–23]. Cephalometric X-rays have been performed to follow patients when performing RME in the past, but cephalometric X-rays have limitations [24,25], and often, as for 3D-CT, not enough attention is given to reproducibility of images over time, and often there is too significant a variability in the technical protocol and head-placement of the child to obtain images that allow a valid comparison of measurements over time. Usage of 3D-CT is not new when considering changes in upper-airway morphology following RME: DiCarlo et al. [26] performed a systematic review after performing a database search using PubMed, Ovid, and Cochrane Library up to December 2016. These authors could only include nine articles in their review that fulfilled their entry criteria of having “solid and coherent protocols when CBCT was adopted to measure airway dimensions and morphology”. And their conclusions were that use of CBCT was inconsistent between studies, and there was too great a heterogeneity between the 3D-CT methodologies used, and no real conclusion could be drawn from the presented data. This conclusion echoes the findings of a similar type of review performed in 2011 by Baratieri et al. [27]. These different reviews of publications performed up to very recent times emphasize the problems associated with 3D-CT: the problem of reproducibility of measurements over time is a real issue, and may be even more important than with cephalometric X-rays. Factors such as timing of respiratory cycle, position of tongue, for example, are elements that must be clearly noted and reproduced when performing studies, in particular follow-up investigations. Collection and analyses must be performed by the same operator.

We built a very rigid protocol, taking into consideration these issues and made great efforts at repeat testing, to include as many elements as possible. When there were any doubts about the critical issues, data were not entered. Collection and analyses must be performed by the same operator. If such efforts are not made, results will have little validity and variability of findings will be shown again between investigators. We found that we had clear orofacial skeletal modifications related to RME, including the changes of the pterygoid processes in our subjects. We did not address the question of the change in volume of the oropharynx associated with the skeletal changes. The validity of these changes is the most controversial issue because different articles report different and often contradictory results. The problem of measurement of volumes in a living individual with regular breathing is a very complex issue, and investigation of respiratory parameters in association with measurements of 3D-CT will most probably be needed. Also, all changes should be measured on an awake subject. Imaging is only a part of a sleep-disordered-breathing investigation, and has to be integrated into the overall results, including those obtained during sleep with nocturnal polysomnography; but the 3D-CT provides valid information on the skeletal changes



A B

Fig. 2. a, b: maxillary width before (a) and after (b) RME.

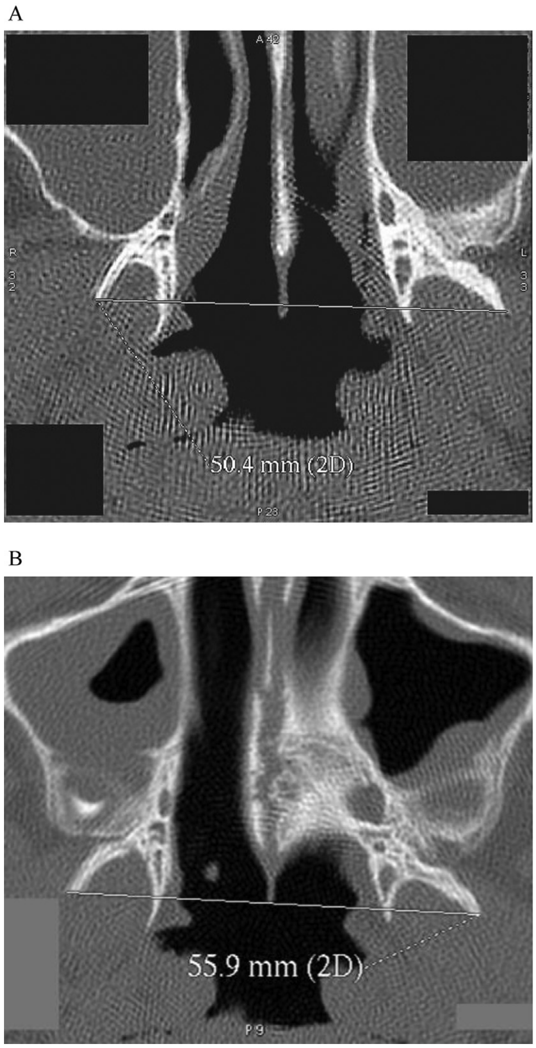


Fig. 3. Pterygoid processes distance before (a) and after (b) RME.

obtained with treatment. Our subjects were part of a longitudinal study and we have already reported the results of the sleep studies that showed an absence of sleep-disordered breathing after RME. Also, the skeletal information is important to follow over time to investigate changes with aging in these growing children.

The issue of the stochastic radiation effect is brought up with the use of CT scanning, but dental CT has greatly improved over time, and the amount of irradiation to which a child is exposed is almost comparable today to that related to cephalometric X-ray exposure, and parents should be made aware of the issue. However, looking at the advantages obtained with dental CT scanning in evaluating children pre- and post-RME, such an approach should be considered in the evaluation of children with OSA as measurements of gain can be clearly defined.

In conclusion, 3D-CT is a technique that may give valid information on the changes obtained with RME on teeth if performed with a valid and rigid protocol when collecting the data. But erroneous collection of data over time is common, and good cephalometric X-rays are better than 3D-CTs obtained without great attention to the technical issues, in particular, complete reproduction of data collection over time. And to date, there is no reliable demonstration of change in UA volumes with longitudinal usage of 3D-CT. This lack of data is related to the absence of agreed-upon protocols for longitudinal studies using 3D-CT and how to select anatomical markers indicative of reproducible images at follow-up studies.

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Conflict of interest

None of the authors has conflict of interest.

We declare no conflict of interest that could be perceived as prejudicing the impartiality of the research reported herein.

The ICMJE Uniform Disclosure Form for Potential Conflicts of Interest associated with this article can be viewed by clicking on the following link: https://doi.org/10.1016/j.sleep.2018.11.023.

References

[1] Pirelli P, Saponara M, Guilleminault C. Rapid maxillary expansion (RME) for pediatric obstructive sleep apnea: a 12-year follow-up. Sleep Med 2015;16:933–5.
[2] Giancotti F, Pirelli P. Rapid expansion of the upper jaw. 1. Diagnosis and indications. Attual Dent 1987;3(20):20–7.
[3] Podesser B, Williams S, Crismani AG, et al. Evaluation of the effects of rapid maxillary expansion in growing children using computer tomography scanning: a pilot study. Eur J Orthod 2007;29:37–44.
[4] Garib DG, Castanha Henriques JF, Janson G, et al. Rapid maxillary expansion-tooth tissue-borne versus tooth-borne expanders: a computed tomography evaluation of dentoskeletal effects. Angle Orthod 2005;75(4):548–57.
[5] Zeng J, Gao X. A prospective CBCT study of upper airway changes after rapid maxillary expansion. Int J Pediatr Otorhinolaryngol 2013;77:1805–10.
[6] El H, Palomo JL. Three-dimensional evaluation of upper airway following rapid maxillary expansion. A CBCT study. Angle Orthod 2014;84:265–73.
[7] Iwasaki T, Saitoh Y, Takemoto Y, et al. Tongue posture improvement and pharyngeal airway enlargement as secondary effects of rapid maxillary expansion: a cone-beam computed tomography study. Am J Orthod Dentofacial Orthop 2013;143:235–45.
[8] Pangrazio-Kulbersh V, Wine P, Haughey M, et al. Cone beam computed tomography evaluation of changes in the naso-maxillary complex associated with two types of maxillary expanders. Angle Orthod 2012;82:448–57.
[9] Guijarro-Martínez R, Swennen GR. Three-dimensional cone beam computed tomography definition of the anatomical subregions of the upper airway: a validation study. Int J Oral Maxillofac Surg 2013;42:1140–9.
[10] Pirelli P, Saponara M, Guilleminault C. Rapid maxillary expansion in children with obstructive sleep apnea syndrome. Sleep 2004;27:761–6.
[11] Villa MP, Rizzoli A, Miano S, et al. Efficacy of rapid maxillary expansion in children with obstructive sleep apnea syndrome: 36 months of follow-up. Sleep Breath 2011;15:179–84.
[12] Fanucci E, Leporace M, Di Costanzo G, et al. Multidetector CT and Dentascan software: dosimetric evaluation and technique improvement. Radiol Med 2006;111:130–8.
[13] Rustemeyer P, Streubühr U, Hohn HP, et al. Low-dosage dental CT. RöFo – Fortschritte auf dem Gebiet der R 1999;171:130–5.
[14] Pirelli P, Addestri S, Tantari C, et al. Digital radiology in dentistry: technical principles, image management, legal aspects, restriction and benefits. Ann Stomatol (Roma) 2004;24:186–90.
[15] Pirelli P, Marullo M, Casagrande M, et al. Espansione rapida del mascellare: effetti sulla funzionalità respiratoria e uditiva. Mondo Ortod 1995;20:129–35.
[16] Pirelli P, Saponara M, De Rosa C, et al. Orthodontics and obstructive sleep apnea in children. Med Clin North Am 2010;94:517–29.
[17] Bhattacharjee R, Kheirandish-Gozal L, Spruyt K, et al. Adenotonsillectomy outcomes in treatment of obstructive sleep apnea in children: a multicenter retrospective study. Am J Respir Crit Care Med 2010;182:676–83.
[18] Huang YS, Guilleminault C. Pediatric sleep apnea and the critical role of oral,facial growth: evidences. Front Neurol 2013;3:184.
[19] Huang YS, Guilleminault C, Lee LA, et al. Treatment outcomes of adenotonsillectomy for children with obstructive sleep apnea: a prospective longitudinal study. Sleep 2014;37:71–6.
[20] Pirelli P, Saponara M, Guilleminault C. Rapid maxillary expansion before and after adenotonsillectomy in children with obstructive sleep apnea. Somnologie 2012;16:125–32.
[21] Guilleminault C, Monteyrol PJ, Huyhn NT, et al. Adenotonsillectomy and rapid maxillary distraction in prepubertal children: a pilot study. Sleep Breath 2011;15:173–7.

[22] Guilleminault C, Huang YS, Quo S, et al. Teenage and sleep-disordered breathing: recurrence of syndrome. Sleep Med 2013;14:37–44.
[23] Camacho M, Chang ET, Song SA, et al. Rapid maxillary expansion for pediatric obstructive sleep apnea: a systematic review and meta-analysis. Laryngoscope 2017;127:1712–9.
[24] Ballanti F, Lione R, Bacetti T, et al. Treatment and posttreatment skeletal effects of R.M.E. investigated with low-dose computed tomography in growing subjects. Am J Orthod Dentofacial Orthop 2010;138:311–7.
[25] Pirelli P, Giancotti A, Pirelli M. ERM: effetti strutturali e ripercussioni sul setto nasale. Mondo Ortod 1996;21:351–60.
[26] Di Carlo G, Saccucci M, Ierardo G, et al. Rapid-maxillary-expansion and upper-airway morphology: a systematic review on the role of cone beam computed tomography. BioMed Res Int 2017:5460429.
[27] Baratieri C C, Alves Jr M, De Souza MMG, et al. Does rapid maxillary expansion have long-term effects on airway dimensions and breathing? Am J Orthod Dentofacial Orthop 2011;140:146–56.