Quality of life of children with sleep-disordered breathing after rapid maxillary expansion: assessment by Osa-18

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ABSTRACT

Objective
This study assessed the quality of life of children with sleep-disordered breathing before and after rapid maxillary expansion.

Methods
A prospective clinical study was done at the University of Brasilia with a sample of 22 children aged 4 to 10 years who complained of difficulty breathing during sleep, snoring, restless sleep and obstructive sleep apnea. Questionnaire administration, clinical tests, nasal endoscopy, and CT scans of the head and neck were done before and after rapid maxillary expansion. The Wilcoxon and chi-square tests, analysis of variance (ANOVA) and odds ratio were used for the statistical analyses.

Results
The quality of life scores improved significantly. The total OSA-18 score decreased from 90.95 at baseline to 46.68 after rapid maxillary expansion. The mean quality of life score increased from 4.5 ± 1.7 to 7.9 ± 1.4, showing a significant improvement in the quality of life of the study children, regardless of the degree of airway obstruction or amount of expansion achieved. All study children complained of snoring, and 68.2% snored every night. After expansion, 36.4% stopped snoring. Additionally, complaints of apnea fell by 77.3%.

Conclusion
Children with sleep-disordered breathing have a poor quality of life mainly because of physical symptoms and sleep disturbances. The quality of life of these children improved significantly after rapid maxillary expansion, regardless of the degree of airway obstruction.


RESUMO

Objetivo
Avaliar a qualidade vida de crianças com distúrbios respiratórios do sono antes e após Expansão Rápida da Maxila.

Métodos
Estudo clínico prospectivo realizado na Universidade de Brasília. A amostra contou com 22 crianças de 4 a 10 anos de idade, com queixas de dificuldade respiratória durante o sono, ronco, sono agitado e apneia obstrutiva do sono. A aplicação dos questionários, os exames clínicos, as tomografias computadorizadas da cabeça e pescoço e as nasofibroscopias foram realizadas antes e depois da Expansão Rápida da Maxila. Os testes Wilcoxon e o qui-quadrado, a análise de variância (ANOVA) e o cálculo da odds ratio foram utilizados para a análise estatística.

Resultados
Houve mudança significativa nos escores de qualidade de vida em todas as comparações dos valores médios, que variaram de 90.95 inicialmente a 46.68 após a Expansão Rápida da Maxila. A média do OSA-18, na escala visual, aumentou de 4.5 ± 1.7 para 7.9 ± 1.4, demonstrando significativa melhora na qualidade de vida das crianças, independente do grau de obstrução adenotonsilar e da quantidade de expansão obtida. Cem por cento da amostra apresentavam queixas de ronco, 68,2% roncavam todas as noites e, após a Expansão Rápida da Maxila, 36,4% não roncavam mais. Já as queixas de apneia diminuíram em 77,3% dos casos.

Conclusão
As crianças com distúrbios respiratórios do sono apresentam baixa qualidade de vida principalmente pelo sofrimento físico e perturbações do sono. A qualidade de vida destas crianças melhorou consideravelmente após a expansão rápida da maxila, independente do grau de obstrução respiratória.


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INTRODUCTION

The expression “sleep-disordered breathing” (SDB) in children refers to a group of respiratory disorders that occur or are exacerbated during sleep. They include snoring, upper airway resistance syndrome (UARS), and obstructive sleep apnea-hypopnea syndrome (OSAHS) in its most severe form.

Snoring is estimated to occur in 8% to 27% of all children, while OSAHS occurs in 2%\(^1\). Yet, the prevalence of OSAHS is believed to be underestimated, especially because of difficult access to accurate and early diagnosis. The golden standard for OSAHS diagnosis is overnight polysomnography, since the clinical history of the patient is not enough to establish the definitive diagnosis\(^2\). Although the incidence of UARS is unknown, it seems to be more prevalent than that of apnea\(^3\).

Nasal obstruction and chronic mouth breathing during childhood are frequently caused by hyperplasia of the pharyngeal and palatine tonsils, which are the main cause of obstructive sleep disorders\(^4\). These hyperplasias may promote many clinical changes ranging from apnea with or without cardiopulmonary repercussions to changes in craniofacial development, postural changes, atypical deglutition, and poor diet\(^5\).

SDB have an important impact on the quality of life of affected children but quality of life improves considerably after surgical treatment\(^6\). However, many children submitted to surgery may experience symptom recurrence during adolescence\(^7\). Rapid maxillary expansion (RME) is a well-known orthodontic procedure. The first orthopedic expansion report, made by Professor Angle in the EUA, dates from 1860. More recently, scientific studies showed the efficiency of RME in the treatment of SDB in children and reported that even better results are achieved when RME is associated with adenotonsillar surgery, regardless of procedure order\(^9\).\(^10\).

Since some children do not have an indication for, refuse to undergo, or cannot undergo adenotonsillectomy, it is important to determine whether RME alone is capable of improving the quality of life of children with SDB.

The objectives of this study were to assess the quality of life of children with SDB after rapid maxillary expansion and compare the results between children with smaller and greater adenotonsillar obstruction and between children who achieved smaller and greater upper airway area at the nasopharynx (UAA), anterior nasal aperture, and intermolar distance.

METHODS

This prospective, non-controlled, before-after-type clinical trial was done at the School of Health Sciences (FS/UnB) and University Hospital of Brasília (HUB) after approval of the local Research Ethics Committee under protocol number 127/08. The convenience sample was selected consecutively from a list of children with indication of adenoidectomy or adenotonsillectomy because of respiratory obstruction diagnosed by the otolaryngology service of the said hospital between August 2009 and August 2010. All children complained of breathing difficulties during their sleep, snoring, restless sleep, and sleep apnea.

Children aged 4 to 10 years of all races and social conditions were eligible for the study after their dental needs were met and their caries treated. However, those with mental disorders or other syndromes, acute or chronic cardiovascular or neuromuscular diseases, and body mass index (BMI) > 25 were excluded. The selected children entered the study after their parents or legal guardians signed a free and informed consent form and the children themselves agreed to participate in the study.

All oral examinations were done by a single orthodontist who classified the children’s occlusions according to Angle’s\(^11\) criteria and checked their lip competence and the presence of unilateral or bilateral crossbite.

The degree of obstruction caused by the palatine tonsils was assessed by nasal endoscopy and their sizes were classified according to Friedman et al.'s\(^12\) staging system as follows: grade I tonsils are hidden within the pillars; grade II tonsils extend to the pillars; grade III tonsils extend beyond the pillars but not to the midline; and grade IV tonsils extend to the midline. Adenoid vegetation was classified as follows: grade I for obstruction of 0% to 25%; grade II for obstruction of 26% to 50%; grade III for obstruction of 51% to 75%; and grade IV for obstruction of 76% to 100%. The groups were then reclassified according to degree of adenoid obstruction and tonsil size. Children with 75% or less of adenoid obstruction were placed in Group A, and those with more than 75% were placed in Group B. Children with tonsil size grade I or II were placed in Group C and those with grade III or IV were placed in Group D.

Tomograms of the head and neck produced by the tomograph GE LightSpeed QXi (Milwaukee, USA) were used for measuring maxillary expansion, given by
the intermolar distance A6 - 6A in centimeters, anterior nasal aperture NA - AN in centimeters (Figure 1), and UAA in square centimeters. The point of reference of the latter was the line on the hard palate connecting the anterior and posterior nasal spine (Figure 2).

Figure 1. Tomograms from top to bottom: anterior nasal aperture (NA-AN) and intermolar distance (A6-6A).

Figure 2. Upper airway area (UAA) at the nasopharynx. The point of reference is the line on the hard palate connecting the anterior and posterior nasal spines. In this image, UAA = 0.952 cm$^2$.

Rapid Maxillary Expansion (RME)

The appliance used for the orthopedic maxillary expansion was based on the prototype developed by Haas$^{13}$, adapted for deciduous and mixed dentitions. It consisted of an acrylic appliance banded with 1.2 mm steel wire with a palatal expansion screw housed in palatal acrylic. The metal structure consisted of the palatine connection bars, welded to the orthodontic bands. The screw, active element of the appliance, was located in the acrylic portion of the appliance, precisely over the palatine raphe, joining the two halves of the expander. As the screw was turned, the palatine widened. In this study, the screw was turned one full turn per day, 2/4 in the morning and 2/4 in the afternoon, until a satisfactory morphology of the upper dental arch was achieved. Overcorrection is always necessary. The activation phase lasted from one to two weeks, depending on the narrowness of the maxillary arch. After this phase, the appliance was allowed to remain passively in the oral cavity for 6 months, the time required for cell rearrangement and stabilization of the results$^{14}$.

Quality of life questionnaire

The OSA-18 questionnaire developed by Franco Jr et al.$^{15}$ and translated into Portuguese, adapted and validated by Silva & Leite$^{5}$ and Lima Jr et al.$^{16}$ was administered to the children’s legal guardians at baseline and at the end of the intervention. The questionnaire consists of 18 items grouped into five domains, namely the child’s sleep disturbances, physical and emotional symptoms, and daytime problems; and the parents’ degree of concern. The frequency in which the problem investigated by the item affects the child is recorded in a scale ranging from 1 to 7. Hence, higher OSA-18 scores reflect a greater frequency and importance of the clinical repercussions of the respiratory obstruction on the quality of life of the child. OSA-18 scores vary from 18 to 126 points. According to Franco Jr et al.$^{15}$, the impact of the respiratory obstruction on the quality of life of patients who score less than 60 points is small. However, its impact on the quality of life of those who score from 60 to 79 points and of those who score more than 80 points is moderate and great, respectively. OSA-18 also has a scale ranging from 0 (zero) to 10 (ten) to record the quality of life of the child according to the reported problems. Zero corresponds to the worst possible and ten to the best possible quality of life.

The head and neck CT scans, nasal endoscopies, and administration of the questionnaires were done at baseline and after RME (T1 and T2). All collected data were stored in the database of the program Statistical Package for the Social Sciences (SPSS), version 17.0 (SPSS Inc., U.S.A). The T1 and T2 variables were compared by the Wilcoxon test. Analysis
of variance (ANOVA) was used for determining the differences between the mean OSA-18 scores in relation to the categorical variables adenotonsillar obstruction, crossbite, occlusion, and age. The chi-square test and odds ratio were also used. The significance level was set at 5% (p<0.05).

RESULTS

The initial study sample consisted of 24 children but one was excluded because of weight gain (BMI increased by 0.5, exceeding 25) and another dropped out. The answers given by 22 children, that is, 14 (63.6%) males and 8 (36.4%) females, and their legal guardians at baseline and after RME to the quality of life questionnaire were assessed. The mean age of the children was 6.0±1.6 years (mean ± standard deviation), varying from 4 to 10 years.

During the intervention, the BMI of 38.1% of the children decreased and that of 61.9% of the children increased. BMI at baseline varied from 11.84 to 20.56, the mean being 15.66. After RME, the mean BMI increased to 16.23, ranging from 13.19 to 22.48. According to the t-test, the difference between the mean BMI before and after the intervention was not significant.

The distribution of the variables sex, age, type of occlusion, presence of crossbite, and lip competence is shown in Table 1. Two (9.1%) children had normal occlusion while all other 20 (90.1%) children had malocclusion.

Table 2 shows the distribution of the sample with respect to the degree of respiratory obstruction at baseline and after the intervention.

| Table 1. Baseline (T1) rate and percentage of the study characteristics. |
|-----------------|-------|-----|
| Characteristic  | Categories | n  | % |
| Sex             | Male       | 14 | 63.6 |
|                 | Female     | 8  | 36.4 |
| Age             | 4          | 3  | 13.6 |
|                 | 5          | 7  | 31.9 |
|                 | 6          | 3  | 13.6 |
|                 | 7          | 4  | 18.2 |
|                 | 8          | 4  | 18.2 |
|                 | 10         | 1  | 4.5 |
| Normal          |            | 2  | 9.1 |
| Occlusion       | Class I malocclusion | 7 | 31.8 |
|                 | Class II malocclusion | 11 | 50.0 |
|                 | Class III malocclusion | 2 | 9.1 |
| Crossbite       | No         | 14 | 63.6 |
|                 | Yes        | 8  | 36.4 |
| Lip competence  | No         | 19 | 86.4 |
|                 | Yes        | 3  | 3.6 |

<table>
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<th>%</th>
<th>T2 N</th>
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<td></td>
<td>Grade II (26 to 50%)</td>
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<td>18.2</td>
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<td></td>
<td>Grade III (51 to 75%)</td>
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<td>9.1</td>
<td>5</td>
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<td></td>
<td>Grade IV (76 to 100%)</td>
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<td>63.7</td>
<td>11</td>
<td>50</td>
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<td>Tonsillar obstruction</td>
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<td>9</td>
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Note: * Not significant

After the reclassification of the children into groups with smaller (A and C) and greater (B and D) obstruction, the results show that patients from Group B (more than 75% of adenoid obstruction) were twice as likely to reach scores below 60 after RME than patients from Group A, that is, children with greater adenoid obstruction experience a greater improvement in their quality of life after RME, but the difference was not significant according to the chi-square test (p=0.375). On the other hand, patients from Group C were 1.25 times more likely to reach scores below 60 after RME than patients from Group D, that is, children with smaller tonsils experienced a greater improvement in quality of life after RME, but again the difference was not significant according to the chi-square test (p=0.806).

The tomograms showed that the mean UAA increased significantly by 0.18 cm², ranging from 1.38 cm² to 1.56 cm² (p<0.05). The mean NA-AN at T1 and T2 were 2.52 cm and 2.71 cm, respectively, that is, there was a significant increase of 0.19 cm; the intermolar distance also increased significantly, with a mean increase of 0.43 cm (p<0.05).

According to the answers given to the first five items of the questionnaire, 100% of the children complained of snoring, some of them sometimes (9.1%), others often (22.7%), and most of them every night (68.2%). After RME, the percentage of children who were snoring often (9.1%) and every night (9.1%) decreased. Twenty-seven percent of the children almost never snored after the intervention and 18.2% snored infrequently. According to the parents, 8 children (36.4%) had stopped
Children with sleep-disordered breathing after RME snoring altogether. With respect to apnea, 81.8% of the children complained of apnea at baseline, but after RME, 77.3% of the children (n=17) reported a reduction in the number of times they stopped breathing or held their breaths during the night.

Table 3 shows the means and standard deviations of the scores of each item of the quality of life questionnaire, the mean score of all the questions, and the mean quality of life score.

![Figure 3](image)

After RME, the sample was again divided into two groups, Group I consisting of children whose quality of life scores were below 60 (n=14, 63.63%) and Group II consisting of children whose quality of life scores were equal to or above 60 (n=8, 36.36%). The mean UAA, anterior nasal apertures, and intermolar distances of these two groups were then compared by ANOVA and the t-test. Although Group I had a greater mean UAA, anterior nasal aperture, and intermolar distance than Group II, the differences were not significant according to ANOVA. However, the expansion achieved by Group I was significantly greater than that of Group II according to the t-test. The patients in Group II (n=8) achieved significant intermolar expansion and anterior nasal aperture increase after the intervention, but UAA did not increase significantly (P=0.176).

The children aged less than 6 years were 1.25 times more likely to achieve quality of life scores below 60 than those aged 7 or more years, that is, younger children experienced a greater improvement in quality of life after RME, but the improvement was not significant according to the chi-square test (p=0.874).

**DISCUSSION**

Earlier studies have already reported the poor quality of life of children with sleep-disordered breathing\(^5,16-17\) because of the distinct repercussions caused by obstruction\(^18\). Thus, the present sample was selected because of their complaints of difficulty breathing during sleep, snoring, restless sleep, and sleep apnea.
The difference between individual baseline and final BMI was not significant. However, the BMI of 61.9% of the children increased after RME. Studies that assessed BMI before and after RME were not found. Silva & Leite\(^5\) did not find a significant difference between BMI before and after adenotonsillectomy.

Only two (9.1%) children had normal occlusion at baseline, while 20 (90.1%) had malocclusion. These findings corroborate the findings of other authors, showing that malocclusion is directly related to respiratory obstruction\(^6,19\). Lofstrand-Tidestrom et al.\(^8\) studied the craniofacial morphology and dental arches of children with respiratory obstruction and found a greater prevalence of crossbite in this group than in children without obstruction. The prevalences of malocclusion (90%) and crossbite (37%) in the present sample are also greater than those in the general pediatric population, which are of 57% and 10%, respectively\(^20\).

Hypertrophy of the adenoids and palatine tonsils is the main risk factor for sleep-disordered breathing in children\(^21\).

All children of the present study presented some degree of adenotonsillar obstruction. At baseline, more than 63% of the children had grade IV adenoid obstruction, and more than 59% had grades III and IV tonsillar obstruction. After RME, only 50% of the children still had grade IV adenoid obstruction. However, percentage of children with different grades of adenoid obstruction (p=0.62) or tonsillar obstruction (p=0.48) did not change significantly after RME. Villa et al.\(^22\) assessed the effect of a mandibular repositioning appliance in children with mandibular retrusion and OSAHS and, contrary to this study, found that 66.7% of their sample experienced smaller tonsillar obstruction after the intervention. This result stemmed from the increase in the lateral and anteroposterior dimensions of the airways in the retropharyngeal space, and RME does not affect this area. Therefore, it is often necessary to treat both arches.

Some studies have excluded children with adenotonsillar hypertrophy because the authors believed the children's SDB would respond poorly or not at all to RME\(^19,23-24\).

In 2004\(^23\), and again in 2005\(^24\), Pirelli et al.\(^19\) published results similar to those of the present study regarding an increase in UAA after RME, and their tomograms also showed greater anterior nasal aperture (1.3±0.3mm) and mean intermolar distance (8.18±0.30mm).

In addition to expanding the maxilla, RME is capable of improving air flow by expanding the nasal cavity. Associated orthodontic tooth movements may also indirectly increase the oropharyngeal space by changing the resting position of the tongue\(^19\).

Many studies have shown the efficacy of RME for treating sleep disorders such as enuresis\(^25\), snoring, and OSAHS, with partial or total resolution of this syndrome\(^5,10,19,23-26\). This finding is similar to those of the present study, even though those studies used different diagnostic methods. In the present study, RME was capable of either reducing the frequency of loud snoring or eliminating the problem in eight (36.4%) children. Complaints of apnea also reduced significantly (p=0.000) according to the Wilcoxon test.

RME is not always capable of improving airflow and nasal resistance. Warren et al.\(^27\) assessed these variables in children aged 10 to 14 years after RME and found that only 45% of them experienced an improvement. Again the authors pointed out that both arches should be treated as needed to get better results in patients with SDB.

Di Francesco et al.\(^18\) believes it is very important to use a detailed and specific questionnaire capable of characterizing sleep and sleep-related disorders and detecting the presence of apnea. Such instrument may often limit, or even replace polysomnography.

A prospective study classified 142 patients aged 2 to 16 years into three groups: allergic rhinitis, adenoid hyperplasia, and adenotonsillar hyperplasia\(^18\). The legal guardians of the children answered a standardized questionnaire about nighttime symptoms. Children with adenotonsillar hyperplasia were younger and experienced snoring, sleep apnea, poor school performance, bruxism, enuresis, and restless sleep more often, and these symptoms were related to apnea, not to allergic rhinitis.

The Wilcoxon test showed that the quality of life of the children in the present study improved significantly (p<0.05) after RME according to the individual scores of the OSA-18 items, the mean global score, and the mean quality of life score (Table 3). This shows that the total score decreased from 90.95, indicating that symptoms had a great impact on quality of life, to 46.68 after RME (Figure 3), showing that the children's quality of life improved. Likewise, the mean quality of life score of all patients increased from 4.5±1.7 to 7.9±1.4, once again demonstrating a significant improvement in quality of life after RME.

A prospective study was done to assess the quality of life of children with a mean age of 5.93 years complaining of restless sleep, apnea, and snoring before and after adenotonsillectomy. The mean OSA-18 score at baseline
was 82.83, and after surgery, 34.15. The total OSA-18 scores and OSA-18 domain scores before and after surgery differed significantly, as observed in the present study. The authors concluded that sleep-disordered breathing has an important impact on the quality of life of children and improves considerably after adenotonsillectomy.

The OSA-18 domains that improved most after RME were “sleep disturbances,” “physical symptoms,” and “caregivers’ concern.” Similarly, Di Francesco et al. used a questionnaire developed by Serres et al. and found a significant improvement in the quality of life of children submitted to adenotonsillectomy, especially in the domains “sleep disturbances” and “physical symptoms.” Therefore, it is possible to state that both RME and adenotonsillectomy improve the quality of life of children with SDB.

In 2009, Carneiro et al. analyzed the effect of adenotonsillectomy on the quality of life of children by interviewing the caregivers and found that adenotonsillectomy improved the quality of life of the children substantially in the long term, especially because they snored less frequently, had fewer tonsillitis episodes, and required less antibiotics.

The changes in the total scores of the “emotional symptoms” and “daytime function” domains were smaller, but still significant. When the items were assessed individually, “mood swings and temper tantrums,” and “discipline problems” did not improve significantly after RME. Even during OSA-18 validation, Franco et al. reported that these two domains, “emotional symptoms” and “daytime function,” were only modestly correlated with OSA-18, given the diversity of factors that affect these domains.

In the “daytime function” domain, the item “poor attention span or concentration” improved significantly after RME (p<0.05). According to Huang et al., obstructive sleep apnea (OSA) is associated with poor attention span, hyperactivity, and aggressiveness. It is likely that as airflow and OSA improved, attention span also improved. However, “daytime sleepiness” and “difficulty getting up in the morning” did not change significantly before and after the intervention. As described earlier, excessive daytime sleepiness is one of the most important clinical signs of OSA in adults but is not as common in children with SDB.

When the children were compared with respect to their adenotonsillar obstruction grade and quality of life, they were not significantly different, that is, all children experienced an improvement in quality of life regardless of their adenoid or tonsil obstruction grade.

Pirelli et al. believe RME is more indicated for patients without adenotonsillar obstruction. On the other hand, the important findings of the present study confirm that RME can be indicated for children with SDB, regardless of adenotonsillar obstruction grade. RME is valid in cases without indication for adenotonsillectomy, when the patients or caregivers refuse surgery, when the patients cannot undergo surgery, or when the symptoms recur after surgery. Guilheminaut et al. concluded that RME is indicated for all cases of SDB because it is a complementary treatment but not less important than adenotonsillectomy. Procedure order is unimportant.

In the light of the modern concepts regarding craniofacial growth and development, RME can be used from deciduous dentition, depending on the maturity of the child and appliance acceptance, to the beginning of adolescence, that is, for as long as separation of the mid-palatal suture is possible. The children in the present sample tolerated the appliance well given that only one of them dropped out of the study during the 6-month intervention. It is important to emphasize that the parents are important for treatment success, because they motivate the children and promote oral hygiene.

Although the OSA-18 questionnaire has been validated for the characterization of sleep and associated disorders, polysomnography is still considered the gold standard for diagnosing such disorders. The impossibility of performing polysomnography tests was one of the limitations of this study, as well as the difficulty of separating the children according to allergies, since allergies play an important role in the genesis of SDB. Finally, this study may encourage new scientific studies with greater sample sizes and other diagnostic methods.

CONCLUSION

The study data show that children with SDB have low quality of life, especially with respect to physical symptoms and sleep disorders, which are of great concern to their parents. The quality of life of these children improves significantly after rapid maxillary expansion, regardless of the severity of their respiratory obstruction.

Collaborators

LPV GONÇALVES participated in the entire creation of the project until the final concept. JP COSTA FILHO helped to adjust the methods, especially with respect to the study of the sleep disorders in children, and write and
review the manuscript. MFS ARAÚJO made adjustments to the methods, performed all the otolaryngology tests, wrote the manuscript and reviewed it. FR BARRA performed all the computed tomography scans, analyzed and interpreted the tomograms, and measured the features of interest in the tomograms. AO TOLEDO supervised the study and participated actively in every step of the project and study, and wrote and reviewed the manuscript.

REFERENCES


