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SLOW MAXILLARY EXPANSION AS THERAPEUTIC ALTERNATIVE TO OBSTRUCTIVE SLEEP APNEA IN CHILDREN WITH DOWN SYNDROME: CASE REPORT

Tatiane Maciel de Carvalho
Master in Temporomandibular Disorders and Orofacial Pain (SL Mandic); Orthodontist (ABO-DF); Professor of the Postgraduate Course in Down Syndrome (IEPSIS); Co-Owner Special Odontologia, Brasilia, Brazil

Andreia Baltazar Dias
Degree in Law (UEL); Postgraduate in Down syndrome (IEPSIS), Londrina, Brazil

Cassio Hideki Higa
General practitioner and Acupuncturist, Londrina, Brazil

Alexandre Franco Miranda
Doctor and Master in Health Sciences (UnB); Professor of the Dentistry course at the Catholic University of Brasília (UCB) – Dentistry for People with Disabilities; Co-Owner Special Odontologia, Brasilia, Brazil

Abstract: The sleep plays key role in human cognitive and weight development. Obstructive sleep apnea syndrome (OSAS) is featured by partial or total obstruction of upper airways during the sleep, associated with oxygen desaturation, fragmentation, and changes in restful sleep stages. It presents multifactorial etiology and concerns individual anatomical, functional and genetic aspects, as well as triggers important cardio-respiratory, cognitive and behavioral diseases. Trisomy 21 (Down syndrome) is featured by anatomical, neuromuscular and neurocognitive changes that lead to higher incidence of breathing disorders during the sleep. The aim of the present study is to report the dental intervention strategy, based on multidisciplinary planning applied to a 3-year-old male patient with T21, who presented restless and fragmented sleep, and apnea hypopnea index (AHI) of 7.2, with 19.6 awakenings per hour and minimum oxygen saturation of 86% overnight, in initial polysomnography requested by a neurologist. Conservative and customized orthodontic treatment based on maxillary disjunction with HAAS-type appliance, three times activation a week - performed by family members after guidance by the dental surgeon in charge - was recommended. A new polysomnography, which showed reduced AHI value (0.2 per hour of sleep) and minimum saturation of 92%, was carried out after 5 months of active orthodontic treatment combined to the follow-up by a speech therapist – this reduction has effectively helped improving the child's quality of sleep. The patient used dental appliance for 4 months without activations, and it allowed the bone remodeling phase. According to the present case report, the conservative orthodontic intervention through slow maxillary expansion, based on multidisciplinary practices and strategies, is an important treatment to improve the quality of sleep of children with Down syndrome and OSAS.
Keywords: Down Syndrome; Orthodontics; Obstructive Sleep Apnea; Quality of life.

INTRODUCTION

Obstructive Sleep Apnea Syndrome (OSAS) is an organic disorder capable of affecting individuals in different age groups. It is featured by obstructive episodes caused by muscle relaxation, and narrowing or interruption of airways, a fact that results in hypoxemia (decreased oxygen saturation in the blood) and hypercapnia (increased carbon monoxide saturation in the blood), as well as contributes to rise heart rate and blood pressure[1]. This change in blood pH, which is quite health harming, results in micro awakenings that help fragmenting the sleep to recover an ideal breathing flow and satisfactory blood conditions[2,3].

Sleep disorders frequency in childhood has strong impact on quality of life, mainly in children suffering with disabilities that interfere with both craniofacial formation and stomatognathic system, such as the Down syndrome. Sleep disorders decrease cognitive function and cause attention and memory deficit. Sleep deprivation, in some cases, leads to hyperactivity and mood swings[4,5].

Down Syndrome (DS) or Trisomy 21 (T21) is the most common chromosomal anomaly in humans; its incidence reaches one case in every six-hundred thousand newborns. This condition leads to individuals' physical and intellectual development delay[1,2,4,6].

Individuals with Down syndrome present changes in both craniofacial formation and stomatognathic system, with emphasis on muscular hypotonia, midface underdevelopment, palate atresia, adenotonsillar hypertrophy and nasopharynx narrowing, which causes inadequate tongue position and predisposition to increased breathing obstruction frequency during the sleep[4,5,7,8].

Multidisciplinary planning and practices involving health professionals and family members in the early diagnosis and treatment of these patients are essential to achieve

beneficial outcomes. The evaluation of children with T21 by dentists must head towards investigating OSAS existence, because, oftentimes, patients present high occurrence indices of it (55%-97%) [8,9,10].

OSAS children's excellency evaluation must encompass the following factors: clinical history and detailed anamnesis, past medical history, application of a specific questionnaire to parents and guardians about quality of sleep, request for essential complementary exams, such as polysomnography, and detailed intra- and extra-oral exmination[10,11].

Bulky adenoids and palatine tonsils are the most prevalent obstructive factors in OSAS children, but some other factors can be associated with Down syndrome. Lack of strategies and multidisciplinary approaches have direct influence on a likely therapeutic failure[12,13,14].

Polysomnography, which is a specific exam carried out in sleep laboratories, is essential, because this is the best and most accurate parameter to find sleep disorders[5,12,15].

After OSAS confirmation, the therapeutic approach must follow the obstruction's etiology and clinical symptoms severity. The orthodontist, which is a dental surgeon trained in craniofacial changes, can conservatively intervene with these patients by recommending the installation of maxillary expander orthopedic appliances[16,17]. The fast and slow maxillary expansion aims at enlarging the nasomaxillary complex to help improving air flow and reducing air passage resistance[18,19].

The aim of the present study was to use a case report to approach the clinical and conservative intervention strategy applied to a 3-year-old patient with Down syndrome. His apnea and hypopnea index was corrected based on multidisciplinary strategies and on the appropriate use of orthodontic appliances for maxillary expansion, since it helps reducing systemic breathing losses.

CASE REPORT

A 3-year-old, leukoderma, Asian descent male child with Down syndrome presented to the dentistry office with his mother for buccal condition evaluation. He complained of restless and unrefreshing sleep, which was confirmed by initial polysomnography required by his neurologist.

Extra-oral examination showed hypotonia of the orbicularis oris muscle, underdevelopment of the middle third of the face, with slight projection of the zygomatic bone, mandibular protrusion with large tongue projection. Intra-oral examination allowed observing deciduous dentition, with agenesis of the lower anterior incisors, atresic and high-arched palate with primate space (characteristic of deciduous dentition) and crowding of upper deciduous teeth. He also presented right posterior crossbite, flaccid and depapilate tongue, absence of negative factors such as dental caries and gingival inflammation.

According to family reports, the child presented extremely restless sleep. He moved a lot over the night and had episodes of sleep sitting-up by his bed.

Based on his mother's report, the otolaryngologist in charge had informed the family about his concern with the size of the adenoids and with the possibility of airway obstruction. However, surgical interventions to remove them were not accepted by the child's parents.

Initial polysomnography results have confirmed the nasal breathing resistance presented by the patient, who was compensating body oxygenation by breathing through the mouth, with restless and fragmented sleep. He presented apnea and hypopnea index (AHI) of 7.2, as well as 19.6 micro awakenings per hour of sleep. Minimum oxygen saturation throughout the exam reached 86% over the night.

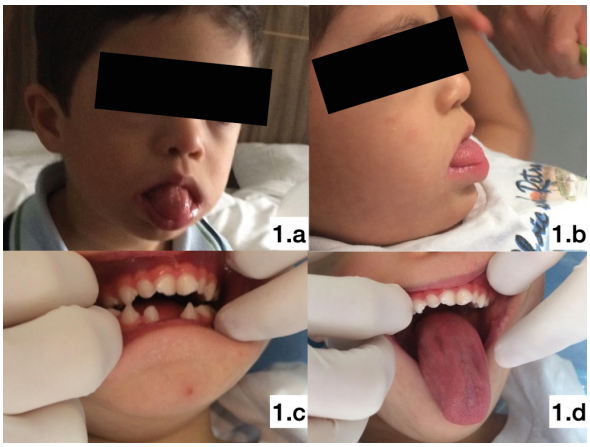

Figure 1 (1a and 1b) – T21 patient profile; it is possible observing little projection of the middle third of the face. Patient presented lack of lip seal, extensive tongue protrusion (Figure 1a– 1b). Intra-oral examination showing crowding of deciduous upper teeth, flaccid and hypotonic tongue. Posterior crossbite on the right side, with agenesis of lower anterior teeth and lack of negative factors, such as caries and periodontal disease (Figure 1b– 1c).

Given this specific case, the use of modified HAAS type circuit breaker was recommended based on detailed craniofacial evaluation, panoramic X-ray, multidisciplinary strategies, signing of the informed consent form and family authorization to conservative orthodontic planning. The adopted protocol was customized to be better adjusted to the patient. This treatment should be combined to speech therapy sessions in order to help better positioning the tongue and lip sealing throughout the treatment.

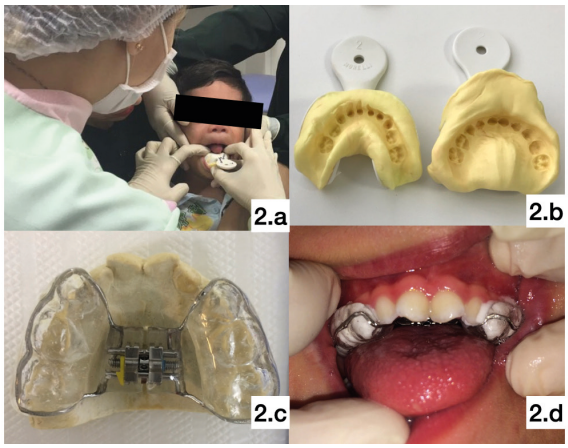

Figure 2 (2a, 2b, 2c and 2d) – Patient modeling with infant tray and condensation silicone. Modified HAAS type appliance recommended for the reported case. Installed HAAS appliance, fixed with ionomeric cement.

Assembling was carried out with size-02 infant tray. Low-flow condensing silicone was selected as molding material, since it makes patient's acceptance easier (Figure 2 – 2a and 2b). The 'talk-show-do' technique was carried out with the aid of a toy, so that the patient could get to know the procedure he would be subjected to. The appliance was manufactured by a laboratory specialized in orthodontic appliances (Figure 2 – 2c). The appliance was fixed on the teeth with glass ionomer cement to allow best oral fixation (Figure 2 – 2d). All hygiene and appliance activation preventive care guidelines were provided to the parents.

Appliance activations were carried out once a week to allow patient adaptation – one full turn in the expander screw was made on a weekly basis. Activations were carried out by the child's parents after orientations and training provided by the dental surgeon in charge. According to family reports, there was no difficulty in activating and cleaning the appliance, or in other care types.

A new polysomnography was conducted after 5 months of active orthodontic treatment combined to speech therapy. It was possible observing drop in the AHI value from 7.2 to

0.2 per hour of sleep and minimum oxygen saturation of 92%, a fact that has contributed to improve the quality of the sleep of the herein assessed child. Intra-oral appliance stabilization was made with fluid resin (Figure 3); the family was recommended to wait for 90 days for likely bone consolidation of the palatal suture. It was also possible observing uncrossed bite after this period-of-time.

According to the mother's report, the follow-up with the otolaryngologist was satisfactory and that this doctor dismissed the surgical intervention after the maxillary expansion carried out by the dentist. The speech therapist also reported patient's better tongue position and speech.

In other words, the specific orthodontic treatment lasted 9 months and it has helped improving this child's quality of sleep and life, as reported.

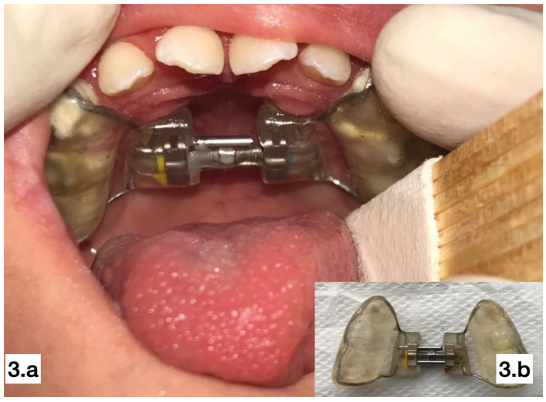

Figure 3a – 3b – Final activations, final screw opening, use of flow resin for stabilization.

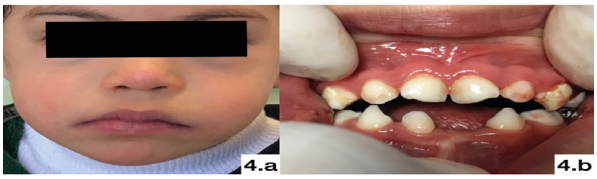

Figure 4 – Occlusion after expander application, uncrossed bite. Significant improvement in lip sealing and tongue posture.

DISCUSSION

The sleep is an essential physiological state necessary for homeostasis, since it keeps the body's physical and intellectual integrity. Thus, it is important assessing the sleep and factors capable of stopping or unbalancing this valuable event for health and quality-of-life promotion[1,2,6].

The Obstructive Sleep Apnea Syndrome (OSAS) can happen at any age[3,5,8,15] and its varying etiology is associated with anatomical, functional, neural and genetic factors that interact with each other for its formation. Nowadays, there is great concern with OSAS early care and detection in children with Down syndrome due to its high prevalence reported in the literature[3,5,7,912].

It is quite clear that children with chromosomal conditions and neuromuscular changes, such as Down syndrome, present high prevalence of infections, breathing obstruction and sleep disorders[4,5,15,18]. The main justifications are related to the fact that craniofacial features observed in children with T21, such as narrow upper airways, maxillary and mandibular hypoplasia, obesity, relative macroglossia, hypertrophy of adenoids and tonsils, and narrow nasopharynx are the main factors leading to OSAS[4,7,8,16,17,18].

OSAS accounts for losses such as attention and memory deficit, it reduces the cognitive function, school performance and daily activities, as well as worsens immunity, helps day-sleep, changes the mood, increases risk of heart and metabolic comorbidities, besides being related to bruxism and restless sleep at night[9,10,11,18].

It is essential having multidisciplinary planning and practices to properly diagnose sleep disorders in children with Down syndrome, because this disease is closely related to health and to individuals' well-being as a whole[6,10,12,14].

It is very important having a guided and detailed anamnesis, with current and past medical history, and the application of a specific questionnaire (Epworth) about quality of sleep, detailed intra- and extra-oral physical examination, image and register sleep-awake exams - such as the golden examination known as polysomnography - to have a successful treatment[3,4,5,7,10,12], as reported in the clinical case.

It is important emphasizing that many children with Down syndrome have a hard time attending the polysomnography exam, because it is expensive, too long, generates discomfort (due to the volume of wire) and is only available in few cities and metropoles[2,3,5].

The oximetry technique and pulse actigraphy with smaller monitors, which are seen as less complex and less accurate – they do not provide reliable results – are the alternatives to polysomnography [1,3,6]. Other sleep diagnostic techniques have been carried out and enhanced to become part of the golden list and standard for the evaluation of children with disabilities, with emphasis on Down syndrome.

The multifactorial nature of obstructive sleep breathing disorders represents a challenge for its treatment. Therapeutic techniques must be taken into account if one has in mind the individual anatomical and clinical changes of different cases, depending on disease severity degree and on risk factors, since these cases demand customized approaches that contribute to maximize the chances of successful treatments[4,14,15,19], as herein reported.

When diagnostic is carried out at younger ages, it is possible choosing less invasive therapeutic options that can become the selected treatment after decision made along with the special child's legal guardians[8,16].

Conservative alternatives such as using intra-oral appliances are available for early and preventive treatments. Maxillary bone expansion in patients with Down syndrome or OSAS provides structural improvements to patients through bone remodeling, since it favors the formation of bigger and wider nasal cavity, which reduces likely breathing resistance, increases air flow and improves tongue position[17,18,19].

The customization of the maxillary expansion protocol must be taken into account. Based on the herein described case, we opted for a slow and gradual treatment to achieve greater patient acceptance and adaptation. It is so, because, in most cases, children with Down syndrome have high buccal sensitivity and feel discomfort in constant appliance use; therefore, they may refuse to adhere to the proposed treatment.

Dental surgeon's adaptation to orthodontic treatment elaboration for children with Down syndrome, with emphasis on OSAS reduction and, consequently, on improving breathing and the life quality of these patients, is a great challenge[16,1718].

CONCLUSION

Conservative orthodontic interventions based on slow maxillary expansion, substantiated by multidisciplinary practices and strategies, are important treatments to improve the quality of the sleep of children with Down syndrome and OSAS, as herein reported.

Individuals with Down syndrome present craniofacial and stomatognathic system features that make this population prone to develop Obstructive Sleep Apnea Syndrome.

Children with Down syndrome need multidisciplinary support encompassing the bio-psycho-social context and focus on these patients' quality of life.

Diagnostic and treatment at younger ages can reduce cardiovascular, neurocognitive and weight sequelae in children with Down syndrome and in the ones who present OSAS diagnostic.

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