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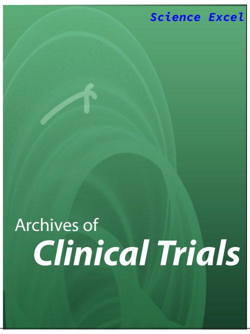

Management Approach on Obstructive Sleep Apnea Pediatric Clinical Case With Myofunctional Therapy

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Keywords

Pediatric Obstructive Sleep Apnea Syndrome, Myofunctional Therapy, Polysomnography, Holistic medical approach

Abbreviations

AAMS: American Academy of Sleep Medicine; A/H: Apnea/Hypopnea; AHI: Apnea/Hypopnea Index; ECG: Electrocardiogram; EMG: Electromyography; EOG: Electrooculogram; LM: Limb Movement; MFT: Myofunctional therapy; NREM: No Rapid Eye Movement; OSA: Obstructive Sleep Apnea Syndrome; OSAHS: Obstructive Sleep Apnea-Hypopnea Syndrome; POSA: Pediatric Obstructive Sleep Apnea Syndrome; PSG: Polysomnography; RDI: Respiratory disturbance index; REM: Rapid Eye Movement; RERA: Respiratory-related arousals

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Abstract

Purpose: *This study aims to review the treatment plan of patients with Pediatric Obstructive Sleep Apnea, as well as to present a clinical case of a pediatric patient with obstructive sleep apnea with improvement of respiratory events through myofunctional therapy treatments.*

Material and Methods: *For the diagnosis of this clinical case, history, intra- and extra-oral evaluation, radiographic examination, and type II polysomnography were performed. After performing 12 months of myofunctional therapy combined with manual physiotherapy, a second type II polysomnography was performed in order to control and register the possible outcomes. Data obtained were evaluated taking into account the normative values of polysomnographic parameters in childhood and adolescence.*

Results: *After therapy, the second polysomnography revealed an improvement on structure of sleep and correction of respiratory events. All events were correct despite the sleep stage. Improvements have been recorded in Apnea/Hypopnea Index, oxygen saturation index and oxygen saturation time <90%. Also, great outcomes on recovering the functional crossbite were recorded and improvement on the behavioral.*

Conclusion: *The new holistic approaches with myofunctional therapies in correlation with postural therapies would appear to be effective in improving the condition of pediatric children with Obstructive Sleep Apnea. It would appear that the use of these therapeutic approaches is most effective in cases of mild Obstructive Sleep Apnea that do not require surgical treatment, as in the treatment of the clinical case just described.*

Introduction

Obstructive Sleep Apnea Syndrome (OSA) is the most prevalent sleep respiratory disorder included in the Sleep-disordered breathing category of ICSD-3 [1]. OSA subjects suffer from episodes of partial or complete upper airway obstruction which causes sleep fragmentation and blood-gas changes (hypoventilation and hypoxemia) [2]. The complete or partial collapse of the upper airway (UA) may be due to intrinsic or extrinsic factors [3], such as anatomic, genetic, or neuromuscular factors [4]. The intrinsic factors are responsible for decreasing the UA patency, and the extrinsic factors, such as fat deposits, hypertrophy of tissues, and certain craniofacial features, cause, instead, an increase in the external pressure of UA [3]. The most common risk factors described in pediatric ages are oral breathing, adenoids and tonsils hypertrophy, posture, and vision plan asymmetry [5].

The incidence of Pediatric Obstructive Sleep Apnea Syndrome (POSA) peaks between 2 to 8 years of age [6]. This happens due to the increased growth of tonsils and adenoids relative to the size of the upper airway in this age group. POSA pathophysiology is complex, which implies a combination of anatomical/ non-anatomical predominant factors [6]. POSA should be addressed by clinical and polysomnographic data collection [6]. In

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children the PSG is done also with capnography[3]. The data collected in PSG allows the diagnosis of OSA and its severity by the determination of the respiratory disturbance index (RDI) [3]. The RDI is the number of respiratory-related arousals (RERA), plus apnea, plus hypopnea events per hour of sleep [3]. RDI scores between 1 and 4.9 events per hour indicate mild OSA, 5 to 9.9 events per hour indicate moderate OSA and more than 9 events per hour indicate severe OSA [3,7]. Children under tha age of 13 should have a RDI bellow 1 respiratory event per hour of sleep.

The severity of OSA determines a wide range of therapy options that consist of both non-surgical and surgical procedures [3]. In pediatric patients, a non-surgical approach may include a therapeutic trial of leukotriene inhibitors [8] and intranasal steroid treatment [4]. The surgical therapeutical approach may be an adenotonsillectomy or partial tonsillectomy [9]. A recent novelty in the treatment of adult and pediatric OSA is myofunctional therapy (MFT) [10]. This entails correct tongue placement, as well as retraining the oropharyngeal structures and oral cavity muscles [10]. MFT consists of isometric and isotonic exercises that work on the oropharyngeal (soft palate, lateral pharyngeal wall) and oral (lip, tongue) structures [10]. Patients perform oral vowel sounds either continuously (isometric exercises) or sporadically (isotonic exercises) as part of soft palate exercises [10,11]. Exercises for the tongue include pushing the entire tongue against the hard and soft palate, moving the tongue along the superior and lateral surfaces of the teeth, and forcing the tongue onto the floor of the mouth [10]. The tongue tip is positioned against the anterior aspect of the hard palate [10]. The buccinators, the lip and the jaw muscles are all addressed by facial exercises [10,11]. Furthermore, patients are instructed to perform specific swallowing and chewing exercises, as well as to inhale nasally and exhale orally both with and without balloon inflation, to address stomatognathic functions [10,11]. Research has demonstrated that physiotherapy and osteopathy therapy are excellent means of achieving a vertebral posture that is well-balanced [12]. Our study aims to review the treatment plan of patients with POSA, as well as to present a clinical case of a pediatric patient with obstructive sleep apnea with improvement of respiratory events through myofunctional therapy treatments. There are few studies on pediatric OSA patients, and there is little information available regarding effectiveness in children.

Materials and Methods

Clinical case background:

Six-year-old male patient, born by cesarean section at 39 weeks and 6 days, presented a clinical suspicion of POSA.

The diagnostic approach was carried out by the following means:

1. Anamnesis and medical history made with the help of the mother.
2. Intraoral/ extraoral clinical examination and postural evaluation.
3. Radiological investigation performed with panoramic and teleradiograph x-rays.
4. Outpatient polysomnography, using EMBLETTA MRP PG/ST+PROXY equipment and recording the following channels: Electrocardiogram (ECG) channels, Electrooculogram (EOG) channels, sub-mentonian and tibial electromyographies (EMGs), nasal pressure, thermistor, plethysmographic bands (thoracic and abdominal), snoring sensor, oximetry, pulse, body position. The analysis of events was carried out manually by differentiated technical professionals and reviewed by doctors with training and experience in polysomnography (double-scoring).

This exam was scored based on the guidelines of the American Academy of Sleep Medicine (AAMS) annual for the Scoring of Sleep and Associated Events: Rules, Terminology and Technical Specifications. Version 2,3 2016 [13].

The final report was based on the Scholle et al. normative values of polysomnographic parameters in childhood and adolescence [14-16].

The first diagnostic examination was conducted in 2021, and the first post-treatment examination was conducted one year later in 2022.

1. The patient's medical history revealed that breastfed (mother refers to left side as child's favorite side) and that sucked the pacifier until 3 years old; also revealed alterations in gripping, sucking, swallowing habits; lingual and paralingual pattern alterations and in addition, the patient presents diction alterations. Has been found out a cross of the mandible to the left side, with lateral inclination of the head to the right and contralateral rotation (Figure 1).
2. The clinical examination revealed a right torsion of the basilar sphenoid synchondrosis.

***Figure 1**. Lateral head inclination*
Age of the patient: 3 years

At postural level: overall postural alteration, with shortening of the muscle and fascial chains to the right of the neck and lengthening of the lateral chains to the left, which cause asymmetry of the ocular plane, shoulder girdle, external rotation of the feet (Figure 2). Presents limited cervical mobility tests for right rotation, compatible with potential right functional torticollis (postural) dysfunction, since the first months of age. In profile examination, accentuated lordosis is observed with superiorization and elevation of the chest and shoulders. The patient has an Asymmetrical Cervical Tonic Reflex to the left with the right side potentially in flexion and the left in extension which had not yet been corrected at the date of the clinical evaluation (Figure 3). Mild limb dysmetria with asymmetrical foot support and mild mid-thoracic sinistro-convex scoliosis. At the secretary, the boy acquires a supine position on the desk (Figures 4a & 4b).

Figure 2. *Pre-treatment posture*
Age of the patient: 8 years

Figure 3. *Asymmetrical Cervical Tonic Reflex*

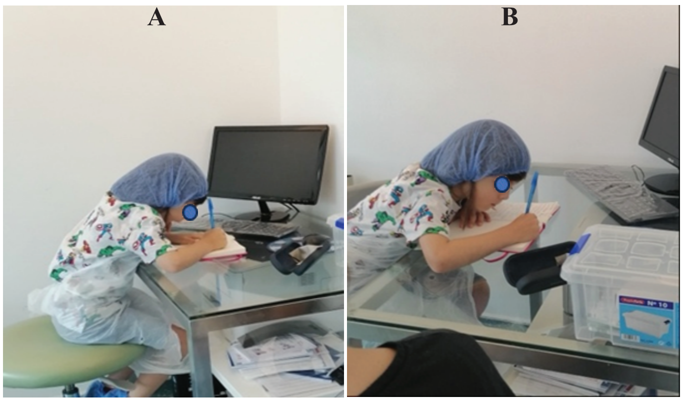

Figure 4. *Child's position to the desk*

Figure 5A. *Pre-treatment intraoral frontal view*

Figure 5B. *Pre-treatment intraoral right lateral view*

Figure 5C. *Pre-treatment intraoral left lateral view*

Figure 5D. *Pre-treatment treatment inferior occlusal view*

Figure 5E. *Pre-treatment upper occlusal view*

At intraoral level, convenience functional lateral crossbite was detected, of the left hemiarch with anterior involvement (Figures. 5a-e). Through mandibular repositioning, dental occlusion between arches is achieved, revealing an end-to-end lateral bite and an anterior open bite (Figure 6). Highly positioned tongue with altered tone as Mallampati IV assessment (Figure 7).

At extraoral level, a significant craniofacial asymmetry with asymmetrical opening pattern of the Temporo Mandibular Joint, Craniometric measurements (Bifrontal, Biparietal, Bizygomatic, Bimastoid and Bigoniac indexes), with examination of the 5 lines, suggestive of potential positional plagiocephaly in infants, with some instability of the Temporo Mandibular Joint and consequent impairment of oral functions (Figure 8).

Figure 6*. Pre-treatment anterior open bite*

Figure 7*. Tongue assessment*

Figure 8*. Pre-treatment extraoral frontal view*

3. From the radiographic analysis, the following emerged:

- Panoramic image revealed a positive overall condition of the oral cavity, with a correct chronology of tooth eruption. (Figure 9).
- Cephalometric analysis reveals straight orthognathic profile, hypertrophy of the soft palate, postural curvature with a decrease in intervertebral spaces at the level of the posterior apophyses of C0, C1, C2 and C4-C5. Hyoid bone with positional rotation to the left (Figure 10).

4. PSG Type II highlighted the following results: The sleep period was 550 min and the efficiency (95,7%) was preserved. There were 14 awakenings and 45 (5.0/h) micro-awakenings, most of which had no specific association. Sleep cyclicity was preserved, with 5 Rapid Eye Movement (REM) periods recorded. REM sleep is normal (17.5%) and the temporal distribution is normal, with a predominance in the second half of the night. The proportions of NREM 1 (2.2%), NREM 2 (49.7%) and NREM 3 (30.5%) were preserved. Deep sleep is preserved (30.5%) and its temporal distribution is typical, with a

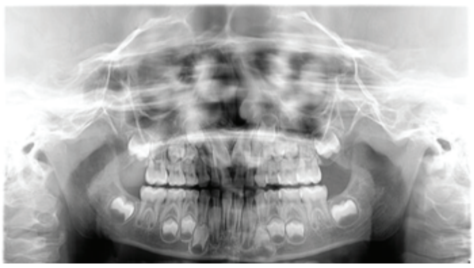

Figure 9*. Pre-treatment panoramic X- Ray*

Figure 10*. Pre-treatment Cephalometric X- Ray*

predominance in the first half of the night (Table 1).

Absence of periodic limb movements (Table 2), but heart rate variations were recorded at an Index of 48,2/h (Table 3). No events of bruxism were reported (Table 2).

An Apnea-Hypopnea Index (AHI) of 1.8/h was found. Around 62,5% of the events recorded were central (Table 4). The average oxygen saturation was 95.7% and was less than 90% for 0.3 minutes (Table 5).

Treatment Plan

Myofunctional therapies carried out for 6 to 12 months, specifically:

Table 1*. Sleep macrostructure*

Sleep period	550 minutes
Sleep efficiency	95,7 %
Number of REM events	2
% NREM 1	2,2%
% NREM 2	49,7%
% NREM 3	30,5%
% REM	17,5%
Number of awakenings	14
Number of micro-awakenings	45
Total Arousals Index	5,0/ h
Arousals associated with Respiratory Events Index	0,3 /h
Arousals without association Index	3,1 /h
Arousals associated with LM Index	1,5/ h

***Table 2**. Movement events during sleep*

LM	3,1 /h
Periodic LMs	0,0 /h
Bruxism (Phasic and Tonic)	0,0 /h
REM	atonia

***Table 3**. ECG/PTT channel assessment*

Autonomous activation Index	48,2 /h
Mean (bpm)	83.8
Standard deviation (bpm)	10,3

***Table 4**. % of the different respiratory events during sleep*

Total number of A/H	**16**	**100%**	**1,8/ h**
Apnea	**10**	**62,5%**	**1,1 /h**
Obstructive	0	0	0,0 /h
Central	10	62,5%	1,1 /h
Mixed	0	0	0,0 /h
Hypopnea (All)	**6**	**37,5%**	**0,7 /h**
RERAS	**0**	**0**	**0,0 /h**
Total number of Obstructive events	**6**	**37,5%**	**0,7 /h**
Total	**16**	**100%**	**1,8 /h**

***Table 5**. Respiratory events during sleep*

Oxygen Desaturation Index	1,5 /h
Average Oxygen Saturation during wake	95,8%
Mean Oxygen Saturation	95,7%
Lowest Oxygen Saturation	89,0%
Saturation time < 90%	0,3 minutes (0.1%)
Average Descent Desaturation	3,5%

***Table 6**. Apnea/hypopnea statistics PSG 2021*

Respiration	Number in REM	REM Index	Number in NREM	NREM Index
Apnea	5	3,1/ h	5	0,7 /h
Obstructive	0	0,0/ h	0	0,0/ h
Central	5	3,1/ h	5	0,7/ h
Mixed	0	0,0/ h	0	0,0/ h
Hypopnea	3	1,9/ h	3	0,4/ h
Obstructive	2	1,2/ h	3	0,3/ h
Central	-	-	-	-
Mixed	-	-	-	-
RDI		5,0/ h		1,1/ h
Total	8	5,0/ h	8	1,1/ h

a) Global postural re-education aimed at correcting the structures identified.

b) Reintegration exercises, with neuromeningeal, vestibular and sensorimotor integration techniques.

c) Manual Therapy aimed at improving the asymmetries described above through techniques to promote better regional circulatory recruitment and normalization of the neuromyofascial alterations of the different muscle groups, the autonomous nervous system, humours techniques, neural techniques, sacro-cranial techniques.

Cranial and epicranial, buccopharyngeal and cervical fascial therapies, with re-education of the upper cervical pattern.

d) Postural and coordinative techniques and exercises that promote the rebalancing of the upper thoracic strait. Synchronization of the five diaphragms, work on the vertebral, sternal and phrenic pericardial ligaments and their relationship with the harmonization of the base and vault peribuccal musculature. Cranial positioning. Neurofascial release of the skull base and upper cervical foramen, with importance in the postural pattern and cervico-thoracic hinge (by the neurovegetative), thoracic and abdominal fascial therapy, work at the level of the thoracolumbar thoracic-lumbar hinge, pillars of the diaphragm and repositioning of the pelvis, as well as the centre of gravity in the base of support (rebalancing the static muscles and muscles and re-education of the curvatures of the spine).

Results

After performing 12 months of myofunctional therapy combined with manual physiotherapy, a second PSG was performed in order to control and register the possible outcomes.

Also, an intraoral evaluation and records were taken in order to compare the evolution of the patient's therapy (Figures 11a-g).

The MTF has helped improve overall stomatognathic function and centered the mandible to the point where it no longer presents a lateral crossbite with mandibular deviation. However, some maxillary compression of the basal bone seems to be observed, so the parents have been recommended to undergo maxillary skeletal expansion, and additionally, vertical control would be also necessary due to the patient's long face.

Improves on the behavioral and school performance were mentioned by parents and schoolteachers.

***Figure 11A**. Post-treatment intraoral frontal view*

***Figure 11B**. Post-treatment intraoral right lateral view*

Figure 11C. *Post-treatment intraoral left lateral view*

Figure 11D. *Post-treatment Cephalometric X- Ray*

Figure 11: E. *Post-treatment frontal view of posture;* ***F***. *Post-treatment profile view of posture;* ***G***. *Post-treatment posterior view of posture*

Table 7. *Sleep macrostructure after treatment*

Sleep period	563,5 minutes
Sleep efficiency	95,3 %
Number of REM events	5
% NREM 1	2,7%
% NREM 2	53,4%
% NREM 3	22,4%
% REM	21,4%
Number of awakenings	8
Number of micro-awakenings	43
Total Arousals Index	4,6/ h
Arousals associated with Respiratory Events Index	0,6 /h
Arousals without association Index	1,9 /h
Arousals associated with LM Index	2,1/ h

Table 8. *Movement events during sleep after treatment*

LM	11,1, /h
Periodic LMs	3,3 /h
Bruxism (Phasic and Tonic)	0,0 /h
REM	atonia

Table 9. *ECG/PTT channel assessment after treatment*

Autonomous activation Index	56,8 /h
Mean (bpm)	74.5
Standard deviation (bpm)	13,5

Table 10. *% of the different respiratory events during sleep after treatment*

Respiratory events	Number	%	Index
Total number of A/H	8	100%	0,9 /h
Apnea	5	62,5%	0,5 /h
Obstructive	0	0	0,0 /h
Central	5	62,5%	0,5 /h
Mixed	0	0	0,0 /h
Hypopnea (All)	3	37,5%	0,3 /h
RERAS	0	0	0,0 /h
Total number of Obstructive events	3	37,5%	0,3 /h
Total	8	100%	0,9 /h

***Table 11**. Respiratory events during sleep after treatment.*

Oxygen Desaturation Index	1,1 /h
Average Oxygen Saturation during wake	96,8%
Mean Oxygen Saturation	96%
Lowest Oxygen Saturation	87,0%
Saturation time < 90%	0,2 minutes (0.1%)
Average Descent Desaturation	3,6%

***Table 12**. Apnea/hypopnea statistics PSG 2022*

Respiration	Number in Rem	REM Index	Number in NREM	NREM Index
Apnea	0	0,0/ h	5	0,7/ h
Obstructive	0	0,0/ h	0	0,0/ h
Central	0	0,0/ h	5	0,7/ h
Mixed	0	0,0/ h	0	0,0/ h
Hypopnea	0	0,0/ h	3	0,4/ h
Obstructive	0	0,0/ h	3	0,4/ h
Central	-	-	-	-
Mixed	-	-	-	-
RDI		0,0/ h		1,1/ h
Total	0	0,0/ h	8	1,1/ h

Discussion

From the evaluation of the PSG results obtained after MFT, the patient, taking into account *the normative values of polysomnographic parameters in childhood and adolescence: arousal events in Sleep Medicine 2012* [16], In the second PSG the respiratory events were corrected. In fact, there was an improvement in the AHI (0.9 /h, Table 10). As in the pre-treatment assessment, 37.5 % of the events recorded were of the obstructive type, but, this time, with a significant reduction in the number of events, with only three Hypopnea events and a total of 8 events if Apnea events are also considered (Table 10), compared with twice as many events obtained in the first examination (Table 4). After treatment, all events taking place in the REM phase (8 A/H events, 5,0/ h Index, Table 6) were corrected (0 A/H events, 0,0/ h Index, Table 12). The multidisciplinary treatment by MFT made it possible to correct all the central apneas from which he suffered (Table 12). MFT`s is especially indicated for patients with a degree of neurological underdevelopment, as is the case with this patient, and for the presence of central apneas.A result that underlines the improvements that treatment with a holistic approach through MFTs can bring to patients with OSA. There was a worsening in the ECG/PTT channel assessment values, with an Index of 56.8 /h, not justified by the respiratory events detected (Table 9), and, therefore, possible indicators of arrhythmias. An Index of 3.3 /h of periodic leg movements was also recorded, possible indicators of metabolic alterations in the child (Table 8). Most of the micro-awakenings occurred during leg movements, but with a large reduction in their number (Table 7). Sleep cyclicity, in both pre- and post-treatment evaluations, was found to be preserved, with 5 REM sleep periods recorded, and, unlike what was obtained in the first analysis, PSG post MFTs revealed that the proportions of phase 1 (2.7 %) and phase 2 (53.4 %), were normal (Table 7). The mean oxygen saturation improved slightly with a value of 96 %, and, an oxygen saturation value below 90 % was found for 0.2 minutes, or 0.0 % of the total analysis time (Table 11). The myofunctional therapy (MFT), previously introduced, has to be conducted by a speech therapist, in which collaboration makes a great advantage in OSAS treatment if performed as combined with the orthodontic therapy [1]. According to multiple studies, MFT provides an adjuvant treatment in OSA therapy [1,10]. this happens trough oxygen saturation increase and orofacial complex myofunctional state improvement [1]. As demonstrated in Camacho's et al. study, myofunctional therapy decreases AHI by approximately 50% in adults and 62% in children [10]. It has been shown in adults, but not in pediatric patients, that lowest oxygen saturation, snoring, and sleepiness outcomes improve in in this group of patients [10]. Values close to what was obtained in the clinic case described below, where there was a 50% improvement in the AHI (0.9/ h post-treatment vs. 1.8/ h in the PSG pre-treatment examination). Furthermore, although in Camacho's et al. work no significant difference in improvements in oxygen saturation values was also demonstrated for the paediatric population, in the clinical case described in our article there were improvements in both the Oxygen Saturation Index (1,5/ h before MFT, 1,1 /h after MFT) and the oxygen saturation time <90% (0,1% before MFT, 0,0% after MFT). Also, in literature, in an article by Guimaraes et al. there is evidence of an improvement in AHI values in patients undergoing MFT [11]. Therefore, MFT could serve as an adjunct to other OSA treatments [10]. Such behavioral modifications provided by MFT can be achieved through daily rehabilitation and re-education exercises [1,17,18]. In a study of Lequeux et. al. particularly in mild OSA cases, physiotherapeutic treatment based on neuromuscular stimulation and musculoskeletal exercises may be useful in the treatment of OSA [19]. In the same study, a several reductions of AHI in the group of patients who had undergone physiotherapeutic treatment with neuromuscular stimulation and muscle exercises when compared to OSA patients who had not undergone physiotherapeutic treatment [19].

Also, rehabilitation exercises are simple to teach, but since parental cooperation is necessary, it's crucial to take the family's psycho-sociocultural level into account in addition to educating and inspiring them [18]. Regrettably, children under 4 years old find it challenging to complete myo-functional exercises because they lack the necessary attention span and consistency to complete them consistently and successfully [2]. These two seem to be some of the limitations or conditions necessary for successful treatment by MFT. According to retrospective studies, children treated with adenotonsillectomy and/or rapid maxillary expansion without orofacial MFT are less likely to achieve long-term OSA remission than those who received adequate orofacial MFT [17]. In the study carried out by Guilleminault et al. a maintenance of results has been demonstrated in reduction of AHI and alleviation of OSA symptoms in patients who continued to perform MFT exercises (follow up of 4 years), as opposed to non-MFT patients who experienced recurrence of pre-treatment OSA symptoms and a worsening of AHI values [17]. Thus, studies show how every child with OSA who has undergone treatment ought to have an oropharyngeal evaluation in order to determine whether neuromuscular rehabilitation could be necessary [18]. There are few studies on pediatric OSA patients, and there is little information available regarding effectiveness in children. So, this might be a topic for further study and a supplement to OSA treatment, as already claimed in the study of Kathryn Gouthro et al [3].

Conclusions

The new holistic approaches with MFTs in correlation with postural therapies would appear to be effective in improving the condition of pediatric children with OSA, especially in cases of mild OSA that do not require surgical treatment. Improvements also occurred in the clinical case just described, which presented exactly mild OSA. A further relevant aspect is the effect of treatment with MTFs in controlling the central apneas from which the patient suffered. No studies have been encountered highlighting this possibility of improvement in central apnoeas through the MTFs and manual therapies approach, and this may also be the starting point for further investigation of this aspect in future studies. However, there remain some limitations to these approaches, such as the correct and constant execution of physiotherapy therapies, MFTs and postural and functional rehabilitation exercises by the young patients, and, therefore, a certain degree of cooperation from the parents would seem to be essential in order to control their correct execution.

Further studies are needed, especially with regard to the maintenance of long-term results by the patients. However, the positive role of MFT as an adjuvant treatment in multidisciplinary treatments for OSA patients seems evident.

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