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REVIEW

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A review on the influence of rapid maxillary expansion and mandibular advancement for treating obstructive sleep apnea in children

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Abstract

This article reviews the orthodontic alternatives for treating pediatric obstructive sleep apnea (OSA). OSA is a multifactorial disease that impairs craniofacial growth and the general health of a developing child and negatively worsens their quality of life. Therefore, it is important to timely diagnose and treat OSA to avoid the progress of the disease, which could otherwise lead to systemic, neurocognitive and social consequences in the patients. In the transverse direction, compression of the maxilla could decrease the diameter of the upper airways and reduce airflow. In the sagittal direction, a retrognathic mandible positioned more posteriorly to the tongue could reduce the available upper airway space and decrease airflow during sleep. Orthopedic treatments for mild to moderate OSA include maxillary expansion using rapid maxillary expansion devices and mandibular advancement using mandibular advancement appliances, which are treatment options only when skeletal discrepancies exist and should be applied after appropriate individual diagnosis for each orthodontic patient. Currently, limited evidence suggests that these therapies could reduce the signs and symptoms and the apnea-hypopnea index (AHI) of OSA.

Keywords

Sleep apnea; Obstructive; Palatal expansion technique; Mandibular advancement; Orthodontics

1. Introduction

Obstructive sleep apnea (OSA) is considered a chronic pathology [1] causing sleep disturbances due to recurrent contraction or collapse in the upper airway. It also causes incomplete restriction of airflow, called hypopnea, or total interruption of airflow for at least 10 seconds during sleep, called apnea [2–5]. The occurrence of OSA in childhood ranges from 1.2% to 5.7% and has a higher prevalence in boys than in girls [6].

Pediatric OSA can cause neurodegenerative pathologies [7], behavioral abnormalities, learning difficulties and growth retardation [8]. It can also negatively impact the quality of life of the patients as it can cause autonomic dysfunction, cardiac arrhythmias, arterial hypertension, remodeling of the ventricular wall and endothelial involvement, whose magnitude depends on the severity of OSA [9–11].

OSA is diagnosed by sleep assessments such as an apnea-hypopnea index (AHI) [11, 12], which is acquired by polysomnography (PSG) and proven to be the best diagnostic method for OSA [13–16].

The treatment options for OSA include continuous positive airway pressure (CPAP), surgery and the use of oral appliances (OA) [17]. Although CPAP is the gold standard treatment [18–20], it is often associated with intolerance and non-compliance in patients [20].

The first-line surgical treatment for patients with OSA is adenotonsillectomy [21, 22]; however, this procedure might be insufficient to solve OSA [23], especially when there is a significant craniofacial anomaly [13, 24]. It was found to be effective in only 25–75% of cases in children [25]. In addition, surgical procedures can lead to severe consequences, such as scarring of the soft palate [3].

OAs can be used as an alternative or adjunctive treatment for OSA patients [26]. Other orthodontic treatment options have also been suggested to decrease mild to moderate OSA symptoms [27]. OAs, including rapid maxillary expansion (RME) and mandibular advancement appliances (MAA), were shown to be valuable alternative treatments in children with OSA related to craniofacial anomalies [28]. The success of OAs in reducing OSA symptoms depends on the enlargement of the airways and reduction in snoring [25].

The objective of this literature review was to analyze and provide an update on the significance of RME and MAA in the treatment of OSA in children.

2. Materials and Methods

An electronic search was performed in PubMed, BVS, Cochrane Library, SciELO, ScienceDirect, Scopus and Google Scholar databases for studies published from January 2011 to November 2021. A highly sensitive search strategy was developed to identify studies of interest using the following keywords: "sleep apnea obstructive", "child", "adult children", "pediatric", "mandibular advancement" and "palatal expansion". Additionally, "AND" and "OR" were used as Boolean operators. The following search formula was constructed for each of the databases using MeSH (Medical Subject Headings) terms: "sleep apnea obstructive" AND "child" OR "adult children" OR "pediatric" AND "mandibular advancement" OR "palatal expansion". Systematic review and meta-analysis, prospective cohort study, literature review and clinical report all included in this review.

3. Results

A total of 170 articles were obtained from the electronic search and the references of the selected studies. We identified 16 articles that discussed orthodontic applications and their relationship with OSA. Information about the authors/year of publication, study type and aim, and conclusions are summarized in Table 1. Most studies focused on decreasing AHI with OAs as a treatment for OSA.

4. Literature Review

4.1 Types of Apneas

Apnea is usually classified as central, obstructive or mixed. In central apnea, the air passage is absent due to insufficient respiratory effort [29]. In obstructive apnea, airflow is not present despite constant respiratory effort due to upper airway obstruction [29]. In mixed apnea, central and obstructive apnea occur successively with no normal breathing between the events [29]. It is important to emphasize that there are differences between pediatric OSA and OSA in adults. For instance, OSA in children can cause behavioral problems, while OSA usually manifests as daytime somnolence in adults [8].

4.2 Signs and symptoms

Some common signs of enlarged tonsils and adenoids include maxillary compression, a thin nasal cavity associated with a deep palate and posterior crossbite [8, 30]. Similarly, an atypical orofacial growth pattern known as adenoid facies was also reported [19]. OSA has been associated with several daytime and nocturnal symptoms [29]. The diurnal ones involve excessive drowsiness and abnormal behaviors such as aggressiveness, hyperactivity, or, in contrast, social isolation and pathological shyness. They may present as repetitive upper respiratory infections and headaches [29]. Comparatively, nocturnal symptoms include enuresis, nightmares, intense sweating, snoring and episodes of apnea that impede the normal sleep cycle and hinder its restorative function [29, 31]. Fatal consequences can occur in the most severe cases of OSA due to cardiorespiratory failure caused by the disease [15].

4.3 Apnea Hypopnea Index

AHI is the main diagnostic criterion of OSA. It is defined as the number of apnea and hypopnea events recorded per hour of sleep [15]. AHI is obtained by PSG examination and is based on the following criteria:

In adults, none/minimal: AHI <5 per hour; mild: AHI ≥ 5, but <15 per hour; moderate: AHI ≥ 15, but <30 per hour, and; severe: AHI ≥ 30 per hour [32]. In children, none: AHI <1 per hour; mild: AHI 1–4 per hour, moderate: AHI 5–9 per hour, and; severe: AHI ≥ 10 per hour [9]. AHI results allow the severity assessment of symptoms, estimate the potential risk of complications over time, and help to guide appropriate treatments [30].

4.4 Diagnostic methods

The diagnosis of OSA is based on the data obtained from laboratory studies, physical examinations, and medical history [11, 29]. The gold standard for diagnosing OSA is PSG [28], consisting of channels for electromyography, electroencephalography, electrocardiography, electrooculography, nasal and oral airflow, chest and abdominal movements, pulse oximetry, carbon dioxide tension and arterial oxygen saturation [14, 29]. PSG provides important data on some parameters related to sleep, including time spent below a certain level of oxygen saturation during the night, number and duration of complete or partial obstructions per hour of sleep, lowest oxygen saturation during each event, presence of arrhythmias, type of heart failure, presence and severity of respiratory disorders, and their impact on the cardiovascular system [29]. It also provides data on the severity of sleep disruptions [29].

4.5 Continuous Positive Airway Pressure

The constant use of CPAP improves symptoms, evidenced by improvement in PSG results in up to 85% of OSA patients. However, potential adverse events include blocked nose, dry mouth and an increased number of awakenings [33].

CPAP consists of using small machines that direct the insufflated air through a tube and a mask placed over the pediatric patient's nose or nose and mouth. Air is then directed towards the back of the pharynx [33] to keep the airway patent and facilitate optimal breathing [8]. However, children may have difficulties adhering to this treatment as they could develop intolerance [15, 26] and non-compliance [34, 35]. Its use in pediatric patients is limited due to concerns associated with growth abnormalities [33] and the risk of developing maxillary retrognathia over time [10, 36]. Some professionals have described the use of forehead-supported masks to mitigate the negative effects of backward pressures on the face [24].

OAs are often preferred over CPAP. However, OAs may cause residual OSA [37], an issue also observed with CPAP therapy, because many patients either reject treatment outright or only partially tolerate it [38].

TABLE 1. Authors table.

Authors	Year	Type of study	Type	Aim	Results
Machado	2016	Systematic review and meta-analysis	RME	To evaluate RME in pediatric patients with obstructive sleep apnea syndrome.	RME in children appeared to be an effective therapy for this syndrome. More randomized clinical trials are needed to determine the success of RME in adults.
Camacho	2016	Systematic review and meta-analysis	RME	To assess data obtained from sleep examinations in pediatric patients with RME.	Improvement in AHI and decreased oxygen saturation were analyzed in children undergoing RME, particularly in the short term.
Sanchez	2019	Systematic review and meta-analysis	RME	To study the results of RME in sleep apnea and hypopnea syndrome and observe the variations in the oximetric variables.	RME seemed efficient for treating mild or moderate hypopnea sleep apnea syndrome, as it improved oximetric parameters. It was efficient as an auxiliary treatment to adenotonsillectomy in severe cases of pediatric patients with maxillary compression.
Yanyan	2019	Systematic review and meta-analysis	MAA	To determine the effects of MAA in pediatric patients with OSA.	MAA can be efficient for mild to severe patients before the end of the pubertal peak. Long-term therapy of at least six months could be more efficient than short-term therapy.
Bahammam	2020	Systematic review and meta-analysis	RME	To examine the main findings on RME in OSA treatment.	AHI improved after RME in pediatric patients with OSA.
Tabrizi	2020	Systematic review and meta-analysis	RME and MAA	To analyze the efficacy of MAA and RME in treating OSA in children.	The results indicated that MAA and RME decreased OSA in pediatric patients.
Jeldez	2020	Meta-analysis	RME	To determine the effects of maxillary expansion on the AHI.	It was not feasible to determine the effectiveness, sleep time, and micro arousals of maxillary expansion on the AHI due to respiratory causes, and existing evidence remains extremely limited.
Droppelmann	2021	Systematic review	RME and MAA	To detail the treatments for OSA in pediatric patients with sagittal or transverse intermaxillary anomalies.	There is not enough evidence to conclude that these devices could completely treat the syndrome, but found that they could reduce AHI and its signs and symptoms.
Nazaralli	2015	Systematic review	MAA	To assess the effectiveness of MAAs for treating OSA in pediatrics.	Although MAAs led to short-term positive effects on AHI scores, it was not possible to conclude whether MAAs were effective in treating OSA in children.
Carvalho	2016	Systematic review	MAA	To analyze the efficacies of OAs in pediatric OSA.	A decrease of at least 50% in the AHI was observed in 9 of the 14 treated subjects.
Koretsi	2018	Systematic review	RME and MAA	To summarize existing evidence from randomized trials on the efficacy of OAs, RMEs, surgically assisted rapid maxillary expansion and MAAs in treating OSA.	OAs effectively lowered AHI, and their use is supported by robust evidence. There was no high-quality research evidence to support therapy with maxillary expansion or MAA in patients with OSA.

TABLE 1. Continued.

Authors	Year	Type of study	Type	Aim	Results
Capalbo	2021	Prospective cohort study	RME	To analyze polysomnographic data and quality of life before and after RME in patients diagnosed with OSA.	RME was efficient in OSA therapy, considering the data obtained from PSG, and improved the quality of life of pediatric patients.
Pirelli	2015	Prospective study	RME	To prospectively assess the long-term effectiveness of RME in a group of pediatric patients with OSA.	A subgroup of OSA children with isolated maxillary narrowing was followed up into adulthood and showed stable long-term results following RME treatment.
Bariani	2021	Literature review	MAA	To study the effects of OAs in correcting mandibular deficiency in OSA therapy.	All studies that used OAs for OSA in pediatric patients showed an improvement in AHI scores.
Galeotti	2016	Clinical report	RME and MAA	Effects of simultaneous palatal expansion and mandibular advancement in a child suffering from OSA.	An improvement in the main respiratory symptoms was observed, while cardiorespiratory sleep assessment revealed a reduction in OSA events
Alexander	2019	Case report	RME	RME followed by adenotonsillectomy was performed for treating OSA.	Interdisciplinary treatment approach yielded significant improvement in sleep and quality of life.

RME: rapid maxillary expansion; AHI: apnea-hypopnea index; MAA: mandibular advancement appliances; OSA: obstructive sleep apnea; OA: oral appliances.

4.6 Surgical treatment

Surgery aims to widen the airway by removing the cause of its obstruction after determining exactly where it occurs [39]. The excess tissue is usually located in the oropharyngeal tract in patients with OSA [39].

Adenotonsillectomy is not only used for children with OSA but is also recommended for patients with adenotonsillar hypertrophy [33]. This is the main anatomical risk factor for OSA [21]. Adentonsillectomy has been shown to reduce the severity of OSA in most children, evidenced by polysomnographic findings and quality of life changes [21]. Persistent residual OSA has been reported after surgery in 25% to 40% of children treated with adenotonsillectomy [33]. Maxillary constriction, mandibular retrusion, a narrow upper airway and a long narrow face are craniofacial morphological characteristics often observed in children who coincidentally have OSA and enlarged tonsils [21].

Uvulopalatopharyngoplasty is a surgical intervention that involves resecting the uvula, part of the soft palate and excess tissues in the oropharynx. It is usually performed simultaneously with tonsillectomy [39]. It is widely used as therapy for OSA in selected patients [39].

Some long-term complications include swallowing difficulties and dry throat. In addition, velopharyngeal insufficiency was also reported in up to one-third of the patients [39]. The success of surgery varies depending on whether it is performed alone (success rate, ~30%) or in conjunction with tonsillectomy (success rate, ~60%) [39].

More extensive surgical approaches can be used in craniofacial disorders when upper airway obstruction is severe such as tracheotomy [40]. Patients requiring this procedure often have neuromuscular disorders leading to hypotonia and severe craniofacial abnormalities [40]. Tracheotomy may also be used as a temporary measure to control severe OSA until another surgical procedure can be performed.

4.7 Oral Appliances

Over the last 10 years, oral appliances have gained increasing recognition as a useful alternative to CPAP [39]. OAs are a common therapy for OSA patients [39]. However, mild to moderate OSA cases can be influenced [31] by non-compliance to CPAP, although CPAP seems more efficient in the definitive treatment of OSA than OAs [6, 41, 42]. There are several factors associated with the therapeutic responses to oral appliance treatment, including differences in devices, treatment protocols and craniofacial and upper airway characteristics [37].

4.8 Rapid maxillary expansion

Candidates for RME are patients with dental crowding, narrow palates or high arches, posterior crossbite, and those with class II or III malocclusions [43]. The objective of RME is to expand the maxilla by separating the middle palatine suture due to the late fusion of its structure [43]. Although this procedure is successfully performed in prepubertal patients, there is great variability in clinical results, mainly in late adolescence and young adults, in whom this treatment could have unpredictable outcomes [43] because chronological age cannot be used as an indicator for determining the developmental stage of the midpalatal suture during growth in these patients [43].

Individual evaluation of the midpalatal suture morphology prior to RME is important. Angelieri *et al.* [43] proposed a methodology for individualized assessment of the midpalatal

suture using cone beam computed tomography (CBCT) [43].

RME is performed in patients in stage A, where the mid-palatal suture is almost a straight high-density sutural line with no or little interdigitation. It is also performed in stage B, where the midpalatal suture assumes an irregular shape and appears as a scalloped high-density line, with less resisting forces and more skeletal effects than when performed during stage C, in which the midpalatal suture appears as 2 parallel, scalloped, high-density lines that are close to each other and separated by small, low-density spaces in the maxillary and palatine bones [43]. On the other hand, it is important to highlight that, despite the greater sutural resistance to conventional RME in stage C, it is still possible to orthopedically widen the maxilla without surgical intervention. Stages A and B are typically observed up to 13 years of age. Comparatively, stage C occurs from 11 to 17 years old, although it may occasionally be observed in younger or older age groups [43].

Fastuca *et al.* [14] used an orthodontic appliance in the non-ossified mid-palatal joint to move the maxillary bones away using lateral pressure [14]. This eliminated the compression and increased the volume of the airway, allowing airflow [44]. Their device also caused expansion and flattening of the palatine arch with an inferior direction of the maxilla and a change in the alignment of the jaw [45]. During RME, there is a reduction in nasal resistance, allowing air through the nose and improving the respiratory condition [45]. RME increases the arch of the upper jaw, which favors the position of the tongue, provides the seal of the lips when the mouth is closed and widens the oropharyngeal space, resulting in a significant reduction in oral breathing [45].

In the study by Fastuca *et al.* [14], the authors reported an increase of ~45% in the transverse nasal area after expansion. However, considering the V-shaped anatomy of the palate of the palatal suture, an increase in respiratory quality as the sole purpose of treatment is not considered an indication for RME [14]. Eichenberger *et al.* [46] reported that in people with normal occlusion, RME could be the last alternative to consider when other therapies have failed or have not shown satisfactory results [46].

Comparatively, Cabrera *et al.* [30] showed a significant improvement in the quality of life of the patients regardless of the severity of their respiratory obstruction and a decrease in OSA symptoms after RME therapy, which were evinced by the results of the PSG [30]. Similarly, Ashok *et al.* [47] concluded that maxillary narrowing was associated with chronic nasal obstruction and that RME could have a key role in alleviating the obstruction [47].

In a systematic review and meta-analysis by Camacho *et al.* [48], the authors found that RME led to a 50% decrease in AHI, producing an improvement in symptoms associated with pediatric OSA following an approximate 3 years of follow-up. Similarly, Bahammam *et al.* [32] reported a 77% reduction in AHI between pre-and post-RME (from 12.05 ± 5.06 to 2.6 ± 1.96) in a follow-up of less than 3 years [32], and after a follow-up ≥3 years, they observed that the improvement in AHI between pre-and post-RME was 73% (from 8.46 ± 7.82 to 3.2 ± 2.62) [32].

Marklud *et al.* [49] discussed the efficiency of OAs over time and expressed that they remained stable for up to 10 years but could be reduced over time [49]. The progression of OSA affects AHI [49]. Efficacy is affected and reduced because patients do not adhere to therapy due to insufficient subjective effects or potential adverse events during OSA follow-up [49]. Pirelli *et al.* [50] prospectively evaluated the effectiveness of RME in OSA pediatric patients using several clinical records of otorhinolaryngology, orthodontics and questionnaire scores annually over a 12-year follow-up period [50].

In a systematic review and meta-analysis by Tabrizi *et al.* [51], the authors assessed AHI before and after RME therapy following the treatment of pediatric OSA. They concluded that the mean difference was 6.37 events/hour (95% CI: 6.02–6.72), which was statistically significant ($p = 0.00$) before and after RME. Sanchez *et al.* [31] conducted a systematic review of the literature and a meta-analysis to analyze the results of RME on AHI and obtained similar data, with a mean reduction of 5.79 events/per hour [31]. Similarly, Camacho *et al.* [48] assessed the data obtained from sleep analysis in pediatric patients who underwent RME for OSA and reported that AHI was reduced from 8.9 events/per hour to 2.7 events/per hour in a follow-up of less than 3 years [48].

4.9 Mandibular advancement

Mandibular advancement has been used for several years to solve malocclusion issues [52] as a therapy for pediatric OSA. There is an increasing number of candidates for MAA with Class II and Division 1 mandibular retrusion malocclusions, which often reflect an imbalance or disharmony between the maxilla and mandible, typically due to the underdevelopment of the mandible and/or overdevelopment of the maxilla; thus, leading to a convex soft tissue profile [53].

The purpose of mandibular advancement is to modify the retrognathic mandibular position and redirect its development toward a more frontal position [28, 53, 54]. In addition, it increases the size of the upper airway through OAs that come in different forms, such as orthopedic, orthodontic, removable, and fixed OAs, thereby reducing the risk factors for OSA [13, 52, 53]. From an orthodontic point of view, they can modify the neuromuscular forces in the craniofacial skeleton and dentition, leading to a series of dentoalveolar and skeletal modifications [18, 25, 29].

The use of OA in patients with skeletal Class II jaw showed an increase in the upper airway dimensions, which persisted even after the cessation of facial growth [30].

MAAs represent an efficient therapeutic option for patients with mild-to-moderate OSA before the age of 13 years [53]. However, a treatment period of at least 6 months before the end of the pubertal growth spurt is required for a noticeable and constant change in mandibular development. The 6-month treatment was shown to have better results than a short-term treatment [53].

The need for mandibular advancement depends on the degree of overjet [52]. A literature review showed that mandibular advancement varies from 3 to 7 mm. When the overjet is reduced, the measurement is recorded by locating the incisors in an edge-to-edge relationship. However, in a larger overjet, the incisor relationship is recorded in two or three stages, bringing the mandible 4 mm forward in each stage and creating

more orthopedic variability, which typically presents a positive change in airway permeability [52].

Regarding the variation in AHI, some studies have shown that regardless of the various treatments and procedures performed, there is a reduction in AHI after therapy. One study suggested that MAAs reduced AHI [52]. In a systematic review by Carvalho *et al.* [29], the authors reported that a clinical trial demonstrated a reduction of at least 50% in AHI in 23 patients with an acrylic resin oral bite plate for mandibular positioning compared with those without treatment [29]. Similar results were obtained in a systematic review by Nazaralli *et al.* [25], in which they suggested that MAAs had positive effects on AHI scores in a short period of time; however, it was not possible to deduce the efficiency of MAAs in the treatment of OSA in children [25].

In a systematic review by Tabrizi *et al.* [51], the effects of Twin Block, Two acrylic plates, Herbst and Modified monobloc were assessed on AHI compared with a control group. They found a mean of −1.79 events/per hour (95% CI: −2.10; −1.48) with a statistically significant difference (p = 0.000) between the two groups; however, the heterogeneity, I^2, was 46%, indicating no significant difference between the two groups (p = 0.16), while the mean difference in AHI effects of MAA before and after OSA treatment in children was 1.84 events/per hour (95% CI 1.60–2.07; p = 0.000) in six studies and the heterogeneity between studies was statistically significant (p = 0.000) [51]. Similarly, Yanyan *et al.* [53] found a significant mean difference in AHI variation for the mandibular advancement group compared to the control group was −1.75 events/per hour (95% CI: −2 .07–1.44; p = 0.00001) [53].

5. Conclusion

Orthopedic treatments for mild to moderate OSA include maxillary expansion using rapid maxillary expansion devices and mandibular advancement using mandibular advancement appliances, which are treatment options only when skeletal discrepancies exist and should be applied after appropriate individual diagnosis for each orthodontic patient. Currently, limited evidence suggests that these therapies could reduce the signs and symptoms and the AHI of OSA. Based on existing literature and evidence, it is not currently possible to conclude the optimal treatment for pediatric OSA as there are few high-quality studies, and existing studies did not have a comparative control group or were limited in terms of small sample sizes, lack of randomization and lack of long-term follow-up results to support the use of RME or MAA in the treatment of patients with OSA.

AUTHOR CONTRIBUTIONS

MVLI—designed the research study. DCAA—performed the research. MVLI, DCAA and LPVL—analyzed the data. DCAA and LPVL—wrote the manuscript. All authors read and approved the final manuscript.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

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