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SYSTEMATIC REVIEW

Effectiveness of mandibular advancement orthodontic appliances with maxillary expansion device in children with obstructive sleep apnea: a systematic review

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Abstract

Background The current review aims to explore the evidence regarding the effectiveness of mandibular advancement orthodontic appliances with maxillary expansion device in treating pediatric Obstructive Sleep Apnea (OSA).

Materials and methods A systematic literature search was conducted across PubMed, Cochrane Central, Web of Science, Embase, Scopus databases, Chinese Biomedical Database, Chinese National Knowledge Infrastructure, and Wanfang. The research involved children and adolescents (under 16 years old) who received mandibular advancement and maxillary expansion functional orthopedic appliances for OSA treatment. We performed narrative reviews and subsequently amalgamated the findings from the studies.

Results Six articles were included for review. Although a small number of studies were included, the research suggested the potential advantages of mandibular advancement for children with OSA. Following treatment, there was a decrease in AHI/RDI, an improvement in sleep quality, and the increase in oxygen saturation.

Conclusions The limited quantity and quality of existing studies necessitate caution when drawing conclusions about the effectiveness of mandibular advancement and maxillary expansion for OSA. In the future, larger and well-designed randomized controlled trials (RCTs) are needed to provide more robust evidence. Patients should be carefully selected, and their orthodontic indications should be thoroughly evaluated before inclusion in such trials. We encourage researchers to design studies that monitor patients over several years to provide a comprehensive understanding of the long-term effectiveness.

Trial registration This study was registered in PROSPERO(CRD42023480407) on November 20, 2023.

Keywords Obstructive sleep apnea (OSA), Mandibular advancement, Maxillary expansion, Children, Adolescent

Introduction

As the most intricate manifestation of sleep-disordered breathing (SDB), the prevalence of pediatric OSA ranges from 1 to 5% [1]. Left unaddressed, it can give rise to a spectrum of symptoms including excessive daytime sleepiness, memory deficits, cognitive dysfunction, nocturnal snoring, nightmares, enuresis, nocturia, disruption of regular metabolic processes, cardiovascular complications, and even psychological disorders [2–4]. Typically,

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adenotonsillar hypertrophy is frequently pinpointed as the primary cause of pediatric OSA [5, 6]. Furthermore, it's important to note that OSA can have long-term impacts on a child's growth and development, including cognitive, behavioral, and cardiovascular consequences. Therefore, timely diagnosis and appropriate treatment are crucial in addressing pediatric OSA to mitigate its potential effects on a child's overall well-being.

Polysomnography (PSG) is widely acknowledged as the gold standard for diagnosing obstructive sleep apnea (OSA) [7]. The severity of OSA is typically evaluated using the respiratory disturbance index (RDI) and the apnea-hypopnea index (AHI). The RDI quantifies the frequency of respiratory arrest, hypoventilation, and respiratory effort related arousal (RERA) episodes per hour of sleep. On the other hand, the apnea hypopnea index (AHI) monitors the number of apnea and hypopnea episodes per hour of sleep. Furthermore, Canto et al. proposed that the pediatric sleep questionnaire (PSQ) has the potential to function as a screening tool and often demonstrates a strong correlation with PSG results [8]. These assessment tools provide clinicians with comprehensive insights into a patient's sleep patterns, aiding in diagnosis and treatment planning.

Fagundes et al. summarized previous studies and reported some craniofacial features in the OSA pediatric group, including "increased total and lower facial height, increased overjet, increased open bite, higher mandible angle, retruded mandible, labial in-competency [9]." Studies have indicated that mandibular retraction plays a significant role in OSA [10], leading to constriction of the upper airway and reduced airflow. Moreover, maxillary stenosis may also result in restricted nasal airflow. To address these anatomical issues, studies have shown that removable functional appliances can be effective in increasing the pharyngeal airway volume in patients with a retrognathic mandible [11]. These appliances work by enhancing the permeability of the upper respiratory tract during sleep, expanding the upper airway, reducing collapse, and subsequently improving the muscle tone of the upper respiratory tract [12]. By utilizing these appliances, clinicians can help optimize the upper airway space and promote better breathing during sleep in individuals with craniofacial abnormalities associated with OSA. This approach offers a non-invasive and potentially effective treatment option for managing pediatric patients with OSA and craniofacial anomalies. Further research is needed to explore the long-term efficacy and potential benefits of this intervention.

Rapid maxillary expansion (RME) has shown promising therapeutic effects in improving upper airway issues in patients with craniofacial developmental abnormalities. RME appears to temporarily increase the volume of the nasal cavity and upper part of the upper airway, while also decreasing nasal airway resistance [9] thereby alleviating nasal passage obstruction [13]. Encouraging therapeutic outcomes of RME in children with maxillary constriction and OSA have been showed. Special consideration was given to the width issue in patients with a retruded mandible. Based on traditional mandibular advancement, a maxillary arch expansion screw was incorporated according to the patient's specific condition, allowing for lateral expansion of the dental arch, thereby promoting not only sagittal coordination of the patient's upper and lower jaws, but also beneficial width coordination. The expanded maxilla provides a prerequisite for mandibular advancement and increases the stability of the orthodontic effect. The primary objective of conducting this review was to comprehensively evaluate the existing scientific data and systematically examine the evidence regarding the efficacy of combining maxillary expansion and mandibular advancement in the treatment of pediatric obstructive sleep apnea (OSA). By assessing the current scientific literature and synthesizing the findings, the researchers aimed to shed light on the extent to which maxillary expansion, when combined with mandibular advancement, can effectively address the challenges posed by pediatric OSA. The review aimed to provide a thorough understanding of the benefits, limitations, and potential risks associated with this treatment approach, ultimately contributing to the improvement of therapeutic strategies for pediatric OSA patients.

Methods

The current study's design adhered to the PRISMA 2020 guidelines [14] and was registered in PROSPERO(CRD42023480407) on November 20, 2023.

The inclusion criteria were established based on the population, intervention, comparison, outcome, study design (PICOS) principle.

- Population (P): Children and adolescents (Under 16 years old) who underwent mandibular advancement functional orthopedic appliances for the treatment of OSA. No gender restrictions applied.
- Intervention (I): Treated with mandibular advancement orthodontic appliance with maxillary expansion device.
- Comparison (C): Self comparison before and after/ Negative controls: untreated group/ Other interventions.
- Outcome (O): The primary outcome was the changes of apnea–hypopnea index (AHI)/ respiratory disturbance index (RDI), and oxygen saturation level.

- Study design (S): Non-randomized trials, cohort and case-control studies were included.

The exclusion criteria comprised:

1) Studies involving adult patients, lacking sleep study data (PSG), or lacking data on the primary outcome.
2) Studies involving syndromic patients or animal.
3) Book or conference abstracts, systematic reviews and meta-analyses.

Information sources

An electronic bibliographic search was performed in the following databases: PubMed, Web of Science, Embase, Cochrane Library, Scopus, Chinese Biomedical Database, Chinese National Knowledge Infrastructure and Wanfang. References from original papers and review articles were cross-checked to identify additional trials. No restrictions were imposed on language or publication date.

Search strategy

Search was performed for articles published until 21st September 2023, (Table S1). Supplementary search as of August 19, 2024.

Selection process

All possibly relevant titles and abstracts were imported into a reference manager (Zotero), and duplicates were removed. Screening was independently conducted by two reviewers (S.Y, J.H), who identified and assessed articles based on the information provided in the title and abstract. References that met the eligibility criteria were included. In cases where an abstract did not offer sufficient information to make a decision, the full text was obtained. If consensus could not be reached, a third reviewer was consulted.

Data collection process

Two independent researchers extracted the data from the included studies, and any discrepancies were resolved through discussion.

Data items

The following data were extracted from each study: general information (author, year of publication); design of the studies; study population (number of patients, age); evaluation method; orthodontic diagnosis and information about the intervention/type of appliance and duration of the treatment. The following outcomes were assessed: AHI, oxygen saturation, RDI before and after the treatment; the improvement of AHI in the treated and control groups. Details are provided in Tables 1, 3 and 4.

Risk of bias

Two reviewers (SY, JH) independently assessed the risk of bias using the Joanna Briggs Institute (JBI) critical appraisal checklist fora) case control、b) cohort study and c) non-randomized controlled trials [15]. In the event of any disagreement, a third reviewer was consulted.

Results

After conducting electronic database searches, 1038 articles were identified and screened for retrieval, and two additional records were identified through other sources, resulting in 699 unduplicated records. Among these, 662 were excluded based on the exclusion criteria during title and abstract screening, leaving 37 articles for full-text review. Ultimately, 6 studies met the inclusion criteria and were selected for qualitative analysis [16–21]. The specific selection process is depicted in Fig. 1.

Study characteristics

The included articles encompassed a time span from 2004 to 2024 with 1 articles in Chinese and 5 articles in English. The studies included four non-randomized controlled prospective study [16, 19–21], one case control study [17], one cohort study [18]. The sample size ranges from 10 person to 94 people, with participants' average ages ranging from 5 to 13.4 years old prior to commencing treatment. Two studies did not report gender ratios [16, 18]. One study only included female patients [20]. The remaining three studies had similar gender ratios [17, 19, 21]. Additionally, five studies reported body mass index (BMI) data [16, 17, 19–21]. Among the included studies, four featured a control group [17–19, 21], the other two compared outcomes before and after treatment. To diagnose and assess the severity of OSA, all studies utilized PSG.

The removable appliances used across the studies included the Modified Twin Block, Herbst appliances, Modified Monoblock, and customized orthodontic appliances such as Sleep Apnea Twin Expander [18]. In one study, the continuous expansion period lasted for 30 days [18], while another study had a continuous expansion period of 15 days [16]. The expansion requirement of a study is to stop expanding when the lingual cusp of the upper molar is opposite to the buccal cusp of the lower molar [21]. In the study of Mastud, all patients underwent upper arch expansion using the Timms protocol (Two turns per day, one in the morning

Table 1 Study characteristics and results

Year—Principal Author	Type of Study	Orthodontic Diagnosis	Type of Treatment	Groups	Sample Size	Age(Year) Sex(M/F) BMI(kg/m^2)	Evaluation	Treatment time	Results
Barker et al. 2023 [15]	NRCT	Full Class II, division 1 malocclusion; mandibular retrognathism; mild transverse maxillary deficiency.	Acrylic-splint Herbst appliance The average expansion time was 15 days. The mean maxillary expansion achieved was 3.19 mm. Mandible was advanced 6 mm. and opened 4 mm vertically. Stepwise activations were completed	/	16	12.6y ± 11.5 months BMI: 18.3 ± 1.8	PSG cephalogram Magnetic Resonance Imaging	12-months	The number of respiratory effort–related arousals (RERAs) and the respiratory disturbance index (RDI)decreased after treatment. There was a reduction in the number of respiratory effort–related arousals (7.06 ± 5.37 to 1.31 ± 1.45 per hour of sleep) due to an increase in airway volume.
Schütz et al. 2011 [16]	Case-control prospective	The treatment group demonstrated a skeletal Class II pattern, with a reduced mandibular length. The OSA patients had slightly narrower inter-tooth distances at all levels measured.	Modified monobloc (full occlusal coverage with maxillary expansion screw and tongue retainer). A Tucat's pearl on a sliding wire was used to determine the reference point for the tip of the tongue.) This custom-made appliance incorporated full occlusal coverage and a central maxillary screw, to allow for accompanying expansion as the mandible was advanced.	Group 1: Treated Group 2: Untreated	-Group 1: 20 -Group 2: 20	-Group 1: 10 M/10F 5.91 ± 1.14y 10M/10F BMI:16.02 ± 3.4 -Group 2: 10 M/10F 6.0 ± 0.71y 10M/10F BMI:20.98 ± 0.48	In-lab PSG (only in group 1) -Epworth sleepiness scale -Lateral cephalogram -Dental measurements on casts	6-months	Significant reduction of AHI. Non-significant change of SaO2. The median AHI score decreased from 7.88 to 3.66. MM reduced daytime sleepiness, and the ESS score decreased from 15.2 ± 4.9 to 7.1 ± 2 after treatment.

Table 1 (continued)

Year—Principal Author	Type of Study	Orthodontic Diagnosis	Type of Treatment	Groups	Sample Size	Age(Year) Sex(M/F) BMI(kg/m^2)	Evaluation	Treatment time	Results
Cozza et al. 2004 [17]	Cohort study	Class II malocclusion (mandible posterior to the maxillary arch)	Upper jaw: Hyrax-type expander with embedded tube Lower jaw: removable acrylic plate connected. with a 0.045 wire Construction bite: mandibular Maximal jumping-Wearing time: night time only (after upper expansion) expansion: 1turn/day for 20 days; 2 turn/day for 10 days	Treated group. Control group: untreated	Treated group: 94 (16 from 6–7 years old; 38 from 7–8 years old and 40 from 8–9 years old) Control group: 113(54 from 6–7 years old; 34 from 7–8 years old and 25 from 8–9 years old)	Treated group: 4.19–7.98y Control group: 6-8.96y	PSG	9 ± 3 months	AHI was significantly reduced in 53% of the treated patient samples, below the pathological threshold (< 1), with a higher proportion in the youngest age group (63%). No positive evolution of respiratory symptoms of this OSAS was observed in the control group. The ODR decreased after the treatment only until the age of 7 years, but not significantly.

Table 1 (continued)

Year—Principal Author	Type of Study	Orthodontic Diagnosis	Type of Treatment	Groups	Sample Size	Age(Year) Sex(M/F) BMI(kg/m^2)	Evaluation	Treatment time	Results
Remy et al. 2022 [18]	NRCT	Class II skeletal malocclusion associated with normal maxilla (SNA, 79° to 84°) and mandibular retrusion (SNB ≤ 76°).	An individual customized twin block was fabricated for each patient. One-step mandibular advancement was performed during wax check-bite recording with an edge-to edge incisor relationship and a 3-mm opening between the maxillary and mandibular incisors. A midline expansion screw was incorporated in the upper part of the appliance if any crossbite or cusp-to-cusp relation of the posterior teeth was noted during bite registration. Maxillary expansion was performed when needed.	Study group: (AHI) > 1.0/h twin-block treatment Control group: (AHI < 1.0/h) the control group received a phase of prejunctional therapy (sectional, fixed orthodontic appliance) to correct occlusal interferences.	- Study Group 34 - Control Group 34	- Study Group 10.29 ± 1.21y 29 M/18F BMI:24.6 ± 2.7 - Control Group 10.42 ± 1.35y 29 M/18F BMI:23.9 ± 2.4	PSG CBCT (cone beam computed tomography)	9 months	At the end of treatment, the AHIs had dropped significantly by 11.2 events/hour ($P < 0.001$).

Table 1 (continued)

Year—Principal Author	Type of Study	Orthodontic Diagnosis	Type of Treatment	Groups	Sample Size	Age(Year) Sex(M/F) BMI(kg/m^2)	Evaluation	Treatment time	Results
Zreaqat et al. 2023 [19]	NRCT	Skeletal Class II due to retrognathic mandible (ANB of > 4°), narrow and constricted maxillary arch, Class II Division 1 malocclusion with full cusp molar relationship, overjet of 5–8 mm	Customized fixed intraoral rapid maxillary expansion with a twin-block mandibular advancement appliance. A bonded upper component consisted of rapid maxillary expander (RME) screw fixed in upper component of twin block and bonded lower component for mandibular advancement. All patients underwent upper arch expansion using the Timms protocol (Two turns per day, one in the morning and one in the evening until the desired expansion was achieved)	Only cases with mild and moderate severity, as indicated by an AHI greater than 5 but less than 29 events/hour, were included in the study	22	11.7 ± 1.5y 10 F BMI:34.12 ± 5.89	PSG lateral cephalograms CBCT	8 months	There was a significant improvement in AHI, events/hour, SpO2%,and sleep efficiency. AHI decreases from 12.18 ± 2.6 to 9.8 ± 2.7 Nadir SpO2 (%) increases from 91.5 ± 8.2 to 97.6 ± 5.9

Table 1 (continued)

Year—Principal Author	Type of Study	Orthodontic Diagnosis	Type of Treatment	Groups	Sample Size	Age(Year) Sex(M/F) BMI(kg/m^2)	Evaluation	Treatment time	Results
Mastud et al. 2024 [20]	NRCT	Class II malocclusion, with retrognathic mandible and narrow maxilla, high palate ANB > 4°,SBN < 78°	Modified Twin-block The sum of the vertical opening distance and the forward extension distance is about 8 mm. The angle between the locking plane of the orthodontic appliance and the He plane is 45 °. A spiral expansion spring is placed at the palatal seam of the upper part of the Twin block functional orthodontic appliance. Stop expanding when the tongue tip of the upper molars is opposite the cheek tip of the lower molars (RME)	Group 1: Adenoid and/ Or tonsillectomy +modified twin-block treatment Group2: twin-block treatment	-Group 1: 10 -Group 2: 11	-Group 1: 10.3 ± 0.95y 5M/5F BMI:15.38 ± 1.71 -Group 2: 10.18 ± 0.98y 5M/6F BMI:15.20 ± 1.58	PSG (ApneaLink Air) cephalogram questionnaire (OSA-18) tonsil examination	Group1: 13.5-months Group 2: 12.4 -months	After treatment, both AHI and OAI values of the two groups of patients decreased, while the lowest blood oxygen saturation increased, and the difference was statistically significant ($P < 0.01$), The total score of two OSA-18 groups decreased. All the change of group1 is greater

PSG Polysomnography, *NRCT* Non-randomized controlled trials, *OAI* Obstructive apnea index,*M* Male, *F* Female.

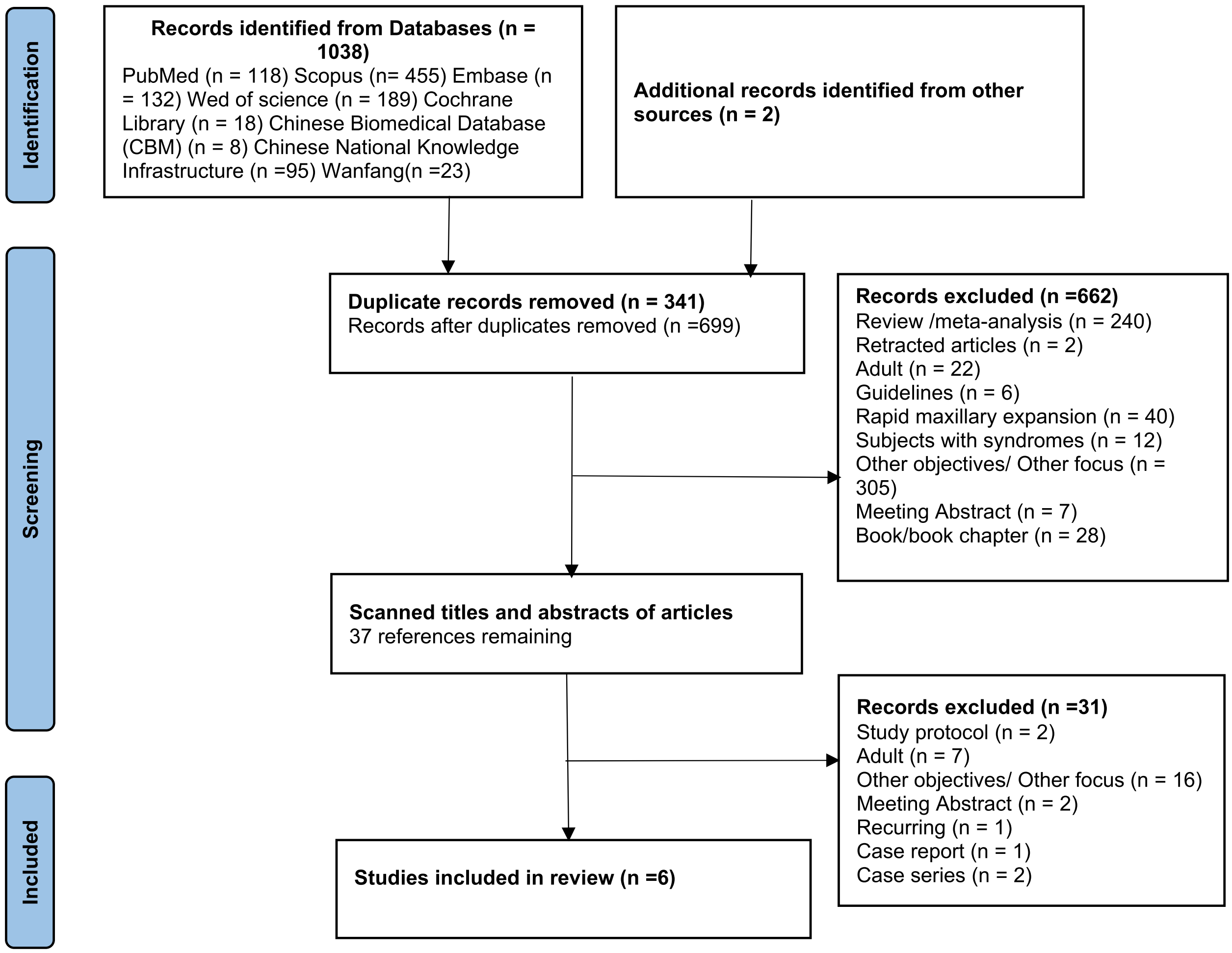

Fig. 1 Flow chart of selection process

and one in the evening until the desired expansion was achieved) [20]. However, two studies did not provide details about the expansion protocol [17, 19]. Furthermore, one study excluded children with adenotonsillar hypertrophy [16]. Three studies conducted tonsillar evaluation [19–21]. One study reported adenoid assessment before treatment and compared the efficacy of adenotonsillectomy and orthodontic combination therapy with orthodontic treatment alone [21].

Risk of bias

In the case-control studies, participant selection bias was evident, and there was no indication of orthodontic indications among the patients undergoing treatment [17]. In two non-randomized controlled trials, no control group was established, and patient follow-up was not reported [16, 20]. For ethical considerations, the cohort study and Zreaqat 's NRCT only gathered pre-treatment data for the control group that had not yet undergone treatment [18]. Two experimental group subjects show overweight BMI, these findings suggest a significant overall risk of bias (Table 2).

Results of individual studies

The heterogeneity in research design and information collection precludes the possibility of conducting a meta-analysis. Consequently, the reported results are descriptive in nature (Tables 3 and 4) In relation to changes in AHI index, a study found that the AHI remained within the normal range both before and after treatment [16]. Additionally, five studies reported a reduction in the AHI index following treatment. One study evaluated the respiratory disorder index (RDI), which exhibited a statistically significant decrease in post-treatment recordings [16]. Various outcomes were assessed in measuring blood oxygen saturation, including minimum oxygen saturation (SaO2), average oxygen saturation, oxygen desaturation rate (ODR) The ODR was defined as the duration during which blood oxygen saturation was ≤96% over the

Table 2 JBI critical appraisal checklist for: a) case control , b) cohort study ,c) non-randomized controlled trials

a) JBI for case control	Cozza et al. 2004 [17]			
1. Were the groups comparable other than the presence of disease in cases or the absence of disease in controls?	Y			
2. Were cases and controls matched appropriately?	N			
3. Were the same criteria used for identification of cases and controls?	N			
4. Was exposure measured in a standard, valid and reliable way?	Y			
5. Was exposure measured in the same way for cases and controls?	Y			
6. Were confounding factors identified?	Y			
7. Were strategies to deal with confounding factors stated?	N			
8. Were outcomes assessed in a standard, valid and reliable way for cases and controls?	Y			
9. Was the exposure period of interest long enough to be meaningful?	NA			
10. Was appropriate statistical analysis used?	Y			
b) JBI for cohort study	Remy et al. 2022 [18]			
1. Were the two groups similar and recruited from the same population?	Y			
2. Were the exposures measured similarly to assign people to both exposed and unexposed groups?	U			
3. Was the exposure measured in a valid and reliable way?	Y			
4. Were confounding factors identified?	U			
5. Were strategies to deal with confounding factors stated?	N			
6. Were the groups/participants free of the outcome at the start of the study (or at the moment of exposure)?	N			
7. Were the outcomes measured in a valid and reliable way?	N			
8. Was the follow up time reported and sufficient to be long enough for outcomes to occur?	Y			
9. Was follow up complete, and if not, were the reasons to loss to follow up described and explored?	Y			
10. Were strategies to address incomplete follow up utilized?	NA			
11. Was appropriate statistical analysis used?	Y			
c) JBI for NRCT (prospective)	YuJiaying 2020 [21]	Schütz et al. 2011 [16]	Zreaqat 2023 [19]	Mastud 2024 [20]
1. Is it clear in the study what is the 'cause' and what is the 'effect' (i.e. there is no confusion about which variable comes first)?	Y	Y	Y	Y
2. Were the participants included in any comparisons similar?	Y	U	Y	N
3. Were the participants included in any comparisons receiving similar treatment/care, other than the exposure or intervention of interest?	Y	N	N	N
4. Was there a control group?	Y	N	Y	N
5. Were there multiple measurements of the outcome both pre and post the intervention/exposure?	Y	Y	Y	Y
6. Was follow up complete and if not, were differences between groups in terms of their follow up adequately described and analyzed?	N	N	N	N
7. Were the outcomes of participants included in any comparisons measured in the same way?	Y	Y	Y	Y
8. Were outcomes measured in a reliable way?	N	N	Y	Y
9. Was appropriate statistical analysis used?	Y	Y	Y	Y

N No, *NA* Not answered, *U* Unclear, *Y* Yes

recorded sleep period. A pediatric OSAS was considered when this rate was exceeded1.4% [18]. Remy's studies evaluated the ODR, reporting no significant changes post-treatment, and in age-group studies, the ODR only marginally decreased prior to the age of 7 [18]. Four studies evaluated SaO2, with Cozza's study demonstrating that orthodontic devices effectively reduced the AHI but had no impact on the minimum arterial oxygen saturation [17]. Similar findings were reported in Schütz's research [16]. However, some studies also noted changes in SaO2. For instance, Mastud's research revealed an increase in SaO2 and a decrease in AHI after treatment, with statistically significant differences in changes before and after treatment [20]. Yu Jiaying's non-randomized controlled trial found that the minimum SaO2 increased in both the experimental and control groups, and the increase was greater in the experimental group with adenotonsillectomy [21]. Mastud et al. found that the lowest SaO2 of patients increased significantly before and after treatment. With regard to quality of life, Cozza et al.

Table 3 Sleep study primary outcomes: AHI/RDI pre- and post-intervention

Year—Principal Author	AHI Initial (Events/h) Mean + SD	AHI Final (Events/h) Mean + SD	Change in AHI (Events/h) Mean + SD	AHI Initial (Events/h) Mean + SD (control group)	AHI Final (Events/h) Mean + SD (control group)	RDI Initial (Events/h) Mean + SD	RDI Intermediate (Events/h) Mean + SD	RDI Final (Events/h) Mean + SD
Schütz et al. 2011 [16]	/	/		/	/	7.3 ± 5.6	4.8 ± 4.2 (P < 0.05)	1.3 ± 1.8 (P < 0.001)
Cozza et al. 2004 [17]	7.88 ± 1.81	3.66 ± 1.70 (P = 0.0003)		/	/	/	/	/
Remy et al. 2022 [18]	6–7:6.9 ± 5.9 7–8:4.4 ± 2.1 8–9:5 ± 3	1.3 ± 1.6 (P < 0.01) 1.1 ± 0.7(P < 0.001) 1.7 ± 1.3(P < 0.001)		6–7:4.5 ± 2.3 7–8:4.7 ± 3 8–9:6.3 ± 5.7	/	/	/	/
Zreaqat 2023 [19]	14.9 ± 5.5	/	11.2 ± 4.6 (P<0.001)	0.4 ± 0.3	/	/	/	/
Mastud 2024 [20]	12.18 ± 2.6	9.8 ± 2.7 (P < 0. 05)		/	/	/	/	/
YuJiaying 2020 [21]	7.15 ± 2.13	1.58 ± 0.80 (P<0.001)		5.87 ± 0.54	3.25 ± 0.50 (P<0.001)	/	/	/

SD Standard deviation, *AHI* Apnea-hypopnea index, *RDI* Respiratory disturbance index

Table 4 Sleep study primary outcomes: oxygen saturation

Year—Principal Author	Lowest SaO2 Initial (%) Mean + SD	Lowest SaO2 Final (%) Mean + SD	Average Sa02 Initial (%) Mean + SD	Average SaO2 Intermediate (%) Mean + SD	Average SaO2 Final (%) Mean + SD	ODR Initial (%) Mean + SD	ODR Final (%) Mean + SD
Schütz et al. 2011 [16]	/	/	97.0 ± 1.1	97.0 ± 1.8	96.8 ± 0.9	/	/
Cozza et al. 2004 [17]	97.39 ± 0.66	96.87 ± 0.85 (P = 0.4072)	/	/	/	/	/
Remy et al. 2022 [18]	/	/	/	/	/	6-7:2.9 ± 2.3 7-8:2.7 ± 1.8 8-9:3.4 ± 3.1	6-7:3.7 ± 4.8 (P = 0.83) 7-8:1.7 ± 1 (P = 0.38) 8-9:2.5 ± 2.2 (P = 0.24)
Zreaqat et al. 2023 [19]	/	/	/	/	/	/	/
Mastud et al. 2024 [20]	91.5 ± 8.2	97.6 ± 5.9 (P = 0.0179)	/	/	/	/	/
YuJiaying 2020 [21]	Group1: 77.30 ± 7.38 Group2: 80.00 ± 5.42	Group 1: 85.20 ± 3.39 (p = 0.009) Group2: 84.73 ± 4.47 (p = 0.000)	/	/	/	/	

ODR Oxygen desaturation rate, *SaO2* Oxygen saturation

evaluated daytime sleepiness symptoms using the Italian version of the Epworth sleep scale (ESS). After treatment, the ESS score decreased from 15.2 ± 4.9 to 7.1 ± 2, indicating a subjective improvement in sleep quality [17]. Yu Jiaying's nonrandomized controlled trial found that after treatment, both groups showed significant improvements in sleep disorders, physical and emotional conditions, daytime function, and the degree of influence on guardians (P < 0.01), and the OSA-18 score in the experimental group decreased more significantly [21].

Discussion

In numerous studies investigating mandibular advancement and maxillary expansion, there is consistent evidence suggesting an improvement in relevant OSA parameters or symptoms, indicating the potential benefits of orthodontic treatment for patients with OSA [16–21]. Schütz's non-randomized controlled trial (NRCT) examined alterations in sleep patterns and craniofacial structure in patients using MM (mandibular advancement) + RME devices [16]. The research revealed a decrease in the frequency of respiratory effort-related arousal (RERA) events and the respiratory disturbance index post-treatment, along with improved breathing, cessation of oral breathing, and the elimination of persistent snoring symptoms. This prospective study involved a small cohort of 16 participants chosen from a pool of 840patients (aged 9 to 14 years) who were being assessed for orthodontic treatment in orthodontic departments. Despite the limited sample size, patients were continuously enrolled. The absence of a control group was deemed appropriate in light of ethical considerations, similar to Mastud's study, but the difference is that the study by Mastud was only conducted on female patients to avoid any bias caused by gender differences. Schütz's study utilized the Herbst device and RME, which are semi-fixed appliances worn continuously (24 h a day) and do not necessitate compliance. Furthermore, the therapeutic impact of Herbst appliances exceeded that of removable appliances within a shorter timeframe. Similar to Schütz's study, in Mastud's research, the orthodontic appliance is a fixed Twin-Block that does not rely on patient compliance. In addition, studies have found that in growth patients with CVM stages 2 and 3, fixed design result in more skeletal effects than movable TB [22]. The present study revealed that the modified twin block effectively increased mandibular growth and led to significant improvement in the posterior airway, decreased AHI, and increased SpO2 levels. Considering the role of RME [9, 13], this study highlights the importance of combining two effective techniques (RME and mandibular advancement with dual dental blocks) to optimize their respective outcomes. One of the limitations of Mastud's study is that it is a single center study. Additionally, the limited sample size of this study limits the application of multivariate analysis. In addition to the small sample size, another constraint of Schütz's study lies in the complexity of the assessment, as 16 patients underwent cephalometry, magnetic resonance imaging, and 4 polysomnography sessions over the 12-month treatment period. The intricate assessments impose a burden on patients and make it challenging to amass data for large sample studies. The study by Zreaqat's demonstrated AHI significantly decreased (by 74.8%), similar to the decrease in Yu Jiaying. However, other studies did not show such a significant decrease (16–18,20). Patient compliance may partially explain these conflicting findings, as subjects who wear dual block appliances more frequently may ultimately have more stable and favorable muscle function to combat upper airway collapse, while non compliant patients do not [19]. In addition, inconsistent patient selection criteria and different etiologies of OSA may also play a role. For example, the difference between the BMI of Mastud's study patients and this study is significant, which may lead to a decrease in the proportion of AHI reduction, only about 19.5%.Another non randomized controlled trial found that patients treated with modified twin block had improved sleep conditions and quality of life, an average decrease of 2.63 times/hour in AHI value, and a 4.73% increase in minimum SaO2, all of which were statistically significant differences [21]. However, in the group undergoing adenotonsillectomy before orthodontic treatment, better therapeutic effects were achieved. We know that a single treatment method cannot be applicable to all patients, and a serialized and personalized treatment plan needs to be developed based on different patients. For patients with moderate to severe OSA accompanied by retrusion and no contraindications for surgery, a multidisciplinary combination therapy of adenoidectomy and orthodontic treatment should be considered. Other studies have also found that AHI values significantly improve after tonsillectomy, and the OSA-18 questionnaire shows significant improvements in quality of life and behavioral problems [23, 24]. Waiting for observation may miss the treatment opportunity or cause more severe symptoms [25].

Cozza's case-control study found that the orthodontic appliance effectively reduced the AHI but had no significant impact on the minimum arterial oxygen saturation [17]. The median AHI score decreased from 7.88 to 3.66 after 6 months of orthodontic therapy. Additionally, the appliance decreased daytime sleepiness and subjectively improved sleep quality, as evidenced by a decrease in the ESS (Epworth Sleepiness Scale) score from 15.2 ± 4.9 to 7.1 ± 2 post-treatment. The gender ratio and age distribution of the sample were similar, but participant exhibited bias, and it was not clarified whether the treated patients met orthodontic criteria. Furthermore, the baseline diagnosis of grade II and/or mandibular recession was not clearly defined. These factors, along with the limited follow-up information and the short-term nature of the treatment (6 months), compromised the validity of the study's findings. Analysis of the lateral cephalograms revealed that the children with OSA demonstrated a skeletal Class II pattern with a shortened mandibular length and a deep overbite. Additionally, the hyoid bone was positioned superiorly in the OSA group. No other significant cephalometric differences were observed between the two groups. While the research suggested that mandibular and rapid maxillary expansion could be an effective treatment for mild to moderate OSA in children, drawing reliable conclusions would necessitate a large sample size and long-term evaluation.

In Remy's subsequent study patients were divided into three distinct age groups, revealing that the reduction in AHI following treatment decreased with advancing age. It was noted that the most favorable treatment period spans the duration of long bone growth until the conclusion of the pubertal growth spurt, with earlier intervention yielding better response. The study observed improved sleep apnea in patients but found that the sleep of patients under 8 years of age remained fragmented or worsened after treatment, as evidenced by an increased pathological Arousal Index (AI) and a decline in sleep quality post-treatment [26]. This decline was attributed to the study itself, as children have numerous sensors throughout their bodies that may affect their sleep quality, suggesting the need to consider this when selecting instruments. Regarding the spontaneous improvement of sleep parameters in children, it remained unclear, and no significant difference in AHI values between the case group and the control group samples was observed. However, the average age of the control group was 15.7 ± 7.6 months older than the case group (mean ± standard deviation). This indicates that AHI did not evolve during the untreated growth process in children, although whether this is linked to a slight age difference necessitates long-term observation.

The sample size in the aforementioned studies is relatively small, ranging from 1 individual to 94 individuals, and details regarding actual patient flow and recruitment are seldom disclosed. Most studies lack stringent inclusion criteria, focusing solely on OSA children with Class II malocclusion, while overlooking the potential for OSA caused by other factors. Furthermore, the suitability for functional correction should consider not only the patient's class II malocclusion, but also their facial contour, occlusion, and compliance. One study, for instance, screened only 16 out of 840 orthodontic patients for inclusion in the trial [16], highlighting the difficulty in identifying suitable candidates based on the inclusion criteria, which has hindered the progress of the study. Moreover, the reproducibility of these findings is limited, as many articles do not utilize dental and maxillofacial examination to select patients for maxillary arch expansion. Some studies even forego cephalometric analysis, potentially leading to the inclusion of unsuitable candidates for arch expansion. The decision to expand the upper airway should not overshadow the indications for arch expansion. While orthodontic diagnosis and treatment typically involve a significant degree of subjective and aesthetic observations, it may be necessary to report basic dental and cephalometric measurements to enhance repeatability. The case-control study by Cozza reported objective dental examination in both the case and control groups prior to intervention, revealing that the distance between mandibular arches in children with OSA was narrower, though it did not address the maxillary arch [17]. A narrow maxilla is often considered a risk factor for OSA in children [27, 28]. However, a parallel randomized controlled trial (RCT) discovered that patients with OSA did not exhibit significant maxillary stenosis when compared to normal reference values. Another study revealed that the maxillary arch of 6-year-old patients with severe OSA in the primary dentition was wider than that of patients with mild to moderate OSA [29]. Additionally, Kim et al. found no correlation between the widening of the nasal maxillary complex and the reduction in AHI [30]. These findings challenge the widespread belief that maxillary width is linearly associated with the severity of obstructive sleep apnea (OSA) in children, underscoring the need for carefully chosen treatment plans and meticulously screened indications for OSA patients.

Furthermore, evidence has demonstrated the efficacy of functional appliances in addressing skeletal Class II malocclusion during adolescent growth spurt [31–33]. However, when utilized before adolescent growth spurt, Class II functional appliances are unlikely to yield clinically significant effects in correcting skeletal relationships. Only three studies included in this article specifically considered this factor when establishing inclusion criteria and included suitable patients based on cervical spine staging [16, 19, 20]. Nonetheless, this appears to contradict Remy's findings that "the earlier the treatment, the better the response."

The primary limitations of this study included the small number of included articles included and the lack of clinical evidence for the majority of the retrieved articles, resulting in generally high bias and small sample size. There is a scarcity of studies on the use of mandibular advancement and maxillary expansion in treating children with OSA, and a lack of randomized controlled evidence to substantiate their benefits, making it challenging to draw definitive conclusions. In the future, larger and well-designed randomized controlled trials (RCTs) will be needed to provide stronger evidence. Future research should aim to include more participants and implement strategies to minimize bias, such as blinding and appropriate randomization. In addition, long-term follow-up after functional therapy is important to evaluate the sustainability of treatment outcomes. Researchers should be encouraged to design studies that monitor patients for many years to comprehensively understand long-term efficacy and potential late-stage side effects.

The primary evaluation measures in this study encompassed changes in AHI/RDI and oxygen saturation. It is evident that twin-block therapy is beneficial in treating pediatric OSA and reducing overall AHIs, although they have not returned to normal pediatric reference values. It is important to note that improvement in OSA can also be gauged through other dimensions, including the snoring index, behavioral changes, neurocognitive impairment, and growth failure, as these factors may serve as the primary motivation for the patient's families to seek treatment. Two studies [34, 35] were excluded from this article due to their reliance solely on sleep questionnaires, without assessing sleep via PSG. Current research has predominantly focused on the individual effects of mandibular advancement (MM) [36, 37] or rapid maxillary expansion (REM) [38–42], with only a limited number of studies exploring the concurrent use of MM + RME. Considering that mandibular retraction often necessitates coordinated width following advancement and maxillary expansion yield superior outcomes compared to individual treatment modalities. Only a few scholars have evaluated the effect of multidisciplinary combination therapy, and the evidence is limited, and more appropriate combination therapy regimens need to be explored in the future [38, 43]. In addition, the soft and hard tissue and aesthetic evaluation after functional orthodontic treatment [44], as well as airway changes, are also targets that can be further studied.

Conclusion

Our systematic review indicates that mandibular advancement appliances with maxillary expansion device can reduce AHI/RDI and increase oxygen saturation in children with OSA. Due to the limited quantity and quality of existing research, caution should be exercised when drawing conclusions on the effectiveness of mandibular advancement and maxillary expansion in treating OSA. In the future, larger scale and well-designed randomized controlled trials (RCTs) will be needed to provide robust evidence. Before undergoing treatment, patients should be carefully selected and their orthodontic indications thoroughly evaluated. We encourage researchers to design studies that monitor patients over several years to provide a comprehensive understanding of the long-term effectiveness and potential late-occurring side effects.

Abbreviations

OSA	Obstructive Sleep Apnea
AHI	Apnea-Hypopnea Index
RDI	Respiratory Disturbance Index
SDB	Sleep-Disordered Breathing
PSG	Polysomnography
RERA	Respiratory Effort Related Arousal
PSQ	Pediatric Sleep Questionnaire
RME	Rapid Maxillary Expansion
HSAT	Home Sleep Apnea Testing
ODR	Oxygen Desaturation Rate
JBI	Joanna Briggs Institute
BMI	Body Mass Index
SaO2	Arterial Oxygen Saturation
OSAS	Obstructive Sleep Apnea Syndrome
ESS	Epworth Sleep Scale
CBCT	Cone-Beam Computed Tomography
NRCT	Non-Randomized Controlled Trial

Supplementary Information

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Supplementary Material 1.

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HJ and YS designed the study. YS and YJ wrote the manuscript. SW, CX and YQ collected, analyzed, and interpreted the data. MH critically reviewed, edited, and approved the manuscript. All authors read and approved the final manuscript.

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All data generated or analysed during this study are included in this published article in the form of tables and figures. Data is provided within the manuscript or supplementary information files.

Declarations

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