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Original article

The contribution of orofacial myofunctional reeducation to the treatment of obstructive sleep apnoea syndrome (OSA): a systematic review of the literature

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ABSTRACT – ***Introduction:*** *Obstructive sleep apnoea syndrome (OSA) is a widespread and under-diagnosed condition, making it a major public health and safety problem. Orofacial myofunctional reeducation (OMR) has been shown to be effective in the multidisciplinary treatment of OSA in children, adolescents and adults and is prescribed at several stages of OSA management.* ***Objectives:*** *The main objective of this systematic literature review was to evaluate the effectiveness of active or passive orofacial myofunctional reeducation (OMR) in the treatment of obstructive sleep apnoea syndrome in children, adolescents and adults.* ***Methods:*** *The systematic literature review was undertaken from the three electronic databases: Medline (via PubMed), Cochrane Library, Web of Science Core Collection, and supplemented by a limited grey literature search (Google Scholar) in order to identify the studies evaluating the effectiveness of the OMR on OSA. The primary outcome of interest was a decrease in the Apnea–Hypopnea Index (AHI) of at least five episodes per hour compared to the baseline state. Secondary outcomes were an improvement in subjective sleep quality, sleep quality measured by night polysomnography and subjectively measured quality of life.* ***Results:*** *Only ten studies met all the inclusion criteria. Eight were randomized controlled clinical trials, one was a prospective cohort study and another was a retrospective cohort study. Six studies were devoted to adult OSA and four to pediatric OSA. All included studies were assessed as "low risk of bias" based on the 12 bias risk criteria of the Cochrane Back Review Group. Based on the available evidence, RMO allows a significant reduction in AHI, up to 90.6% in children and up to 92.06% in adults. It significantly reduces the intensity and frequency of snoring, helps reduce daytime sleepiness, limits the recurrence of OSA symptoms after adenoamygdalectomy in children and improves adherence to PPC therapy. Passive RMO, with the assistance provided to the patient by wearing a custom orthosis, increases adherence to reeducation, significantly improves snoring intensity, AHI and significantly increases the upper airway.* ***Conclusions:*** *Published data show that orofacial myofunctional rééducation is effective in the multidisciplinary treatment of OSA in children, adolescents and adults and should be widely prescribed at several stages of OSA management. Passive RMO, with the pearl mandibular advancement orthosis designed by Michèle Hervy-Auboiron, helps to compensate for the frequent non-compliance observed during active RMO treatments.treatment.*

1. Background

Obstructive sleep apnoea syndrome (OSAS) is a widespread condition characterised by anatomical and/or functional collapse of the upper airways (UA) leading to reduced (hypopnoea) or arrested (apnoea) airflow, oxygen desaturation and fragmented sleep, accompanied by respiratory effort[90,106]. Most patients with OSAS remain undiagnosed110, making OSAS a major public health and safety issue. In children, the many clinical symptoms of OSA have led to the individualisation of two phenotypes, childhood OSAS proper and adolescent OSAS[31].

Untreated childhood OSAS can lead to cognitive impairment[1,126] which appears to be irreversible[12], behavioural problems[94,126], growth retardation[14,24], cardiovascular[38,111] and metabolic complications[51,57].

If left untreated, adolescent OSAS can also cause a wide range of cognitive and behavioural problems[45], from attention disorders[107] to depression, sometimes with risk-taking behaviour and suicidal tendencies[128], cardiovascular[129] and metabolic complications[93].

Untreated adult OSAS is associated with an increased risk of health problems, including cardiovascular disease[49,80,99], carbohydrate-lipid metabolism disorders[16,40] and cancers[86,91,109]. Drowsiness and impaired alertness associated with untreated OSAS have also been shown to increase the risk of work-related injuries and road traffic accidents[47,105], in relation to the fragmentation of sleep induced by abnormal breathing events.

Orofacial myofunctional reeducation (OMR) has been shown to be effective in the multidisciplinary treatment of OSAS in children, adolescents and adults and is prescribed at several stages of these treatments.

The treatment[37,64] of OSAS in children and adolescents is based on a multidisciplinary assessment, which makes it possible to define the therapeutic management, surgical and medical, adapted to each patient. C. Guilleminault[54,64,76] and J. Talmant[119,120] have drawn the attention of the medical world to the imperative need for early diagnosis of OSAS in children and adolescents in order to institute targeted multifactorial treatments and prevent long-term morbidity.

Adeno-tonsillectomy[44,98] is the first-line surgical treatment in children. In children and adolescents, turbinoplasty[139] by laser or radiofrequency may also be indicated and more rarely septoplasty[66], lingual tonsillectomy[71], craniofacial surgery[87] and hypoglossal nerve stimulation[17].

Non-surgical treatment of children, and more specifically adolescents, includes the treatment5 of overweight or obesity through dietary and psychological management[6], combined with increased physical activity and a healthy sleep-wake rhythm. Inflammatory and allergic pathologies are an indication for the prescription of anti-infectious and anti-inflammatory treatments[74], corticoids and anti-leukotrienes. Maxillofacial abnormalities, frequently associated with OSA, will benefit from appropriate treatment[39], rapid maxillary disjunction[19] in the presence of transverse maxillary insufficiency, oral appliances and functional orthopaedic appliances in the case of mandibular retrognathia in a growing patient[23], and active or passive orofacial myofunctional reeducation (OMR) which accompanies any orthodontic treatment or management of temporomandibular dysfunction (TMD)[3]. OMR may also be prescribed independently of orthodontic treatment as an additional medical treatment[18,53], especially after tonsillectomy[134]. The use of continuous positive airway pressure[92] (CPAP) is reserved for severe forms of OSAS.

Orofacial myofunctional reeducation (OMR) is also prescribed for the multidisciplinary management of OSA in adults.

In adults, the most effective and widely prescribed non-surgical gold standard treatment is continuous positive airway pressure (CPAP). In cases of poor compliance or intolerance to CPAP[136], the oral mandibular advancement device (OMAD) is an alternative whose efficacy is comparable to that of CPAP and is attributed to a higher adherence to OMAD than to CPAP[108]. OMAD is also indicated as first-line therapy for moderate OSAS without associated severe cardiovascular comorbidities[58]. If necessary, a behavioural approach is proposed with the introduction of a suitable diet, a physical activity programme[22], reduction of sleeping pills, alcoholic beverages and tobacco consumption[122], and the use of an anti-decubitus device in case of positional OSA[89]. Myofunctional reeducation (MFR), either active or passive[63], is also prescribed and has been evaluated[18,73]. It contributes to improved quality of life[35], reduced snoring[65] and adherence to CPAP[36]. Only one recent pharmacological treatment appears to have been shown to be effective[81,121].

The surgical treatment of adult OSAS[112] involves a number of proposals, including soft tissue volume reduction by tonsillectomy, which is effective in carefully selected patients[21]. Nasal patency can be optimised by nasal valve surgery, septoplasty and turbinoplasty[139]. Hypoglossal nerve stimulation surgery is aimed at contraction of the genioglossus muscle, the main dilator muscle of the pharynx, to suppress pharyngeal collapse during sleep[30]. The highest success rate remains that obtained by maxillomandibular advancement surgery, the only real curative treatment[68].

As the dilator muscles of the pharynx play a key role in maintaining upper airway patency during sleep, specific muscle training methods[18] and exercises using various musical instruments[135], including the didgeridoo[97], have been proposed to treat OSAS.

Following Blandin's[13] recognition in 1836 of the role played by muscle pressures on the shape of the dental arches and his statement of the concept of muscle balance, Rogers[102] emphasised the importance of functional balance, including proper tongue placement, and the therapeutic contribution of OMR to achieving the goals of optimal mandibular growth, facial appearance and nasal ventilation. Since then, there have been countless treatment proposals[95] targeting the structures of the face, oral cavity and oropharynx[8,25,26,42,48,52,55,70,100,103,115,118,127,137].

OMR procedures include isotonic and isometric exercises targeting the oral (lips, tongue) and oropharyngeal (soft palate, lateral pharyngeal walls) structures combined with specific nasal inspiration/oral exhalation, swallowing and chewing exercises. Their most complete description was published by Guimaraes, *et al.*[56].

Numerous studies have shown the effectiveness of orofacial myofunctional reeducation (OMR) in reducing the severity of OSA and associated symptoms in adults[56]. Studies have also shown its effectiveness in reducing snoring[65], improving quality of life[35], improving adherence to CPAP[36], and treating residual OSA after adenotonsillectomy in children[53,134].

Active OMR requires significant cooperation from the patient and, for children, constant involvement of the family in the procedure. To overcome the frequent lack of compliance, authors have proposed the use of passive OMR assisted by a custom-made device[29,63] or prefabricated functional appliances[77,78,101,130,138,140], with for the latter the additional objective of modifying the shape and relationship of the dental arches. The inclusion of these appliances in OMR procedures requires the practitioner to undertake rigorous monitoring to continuously assess the potential adverse effects of the appliances on the dental arches, particularly the vestibuloversion of the mandibular incisors.

The number of studies evaluating the effects of OMR in OSA patients is increasing[35,53,56,114,117] and narrative reviews[83,113], systematic reviews[34,72,73,88,131] and meta-analyses[2,18,20] are regularly published.

2. Objectives

The main objective of this systematic review of the literature was to evaluate the effectiveness of active and passive orofacial myofunctional reeducation (OMR) for the treatment of obstructive sleep apnoea syndrome in children, adolescents and adults. The difference between this systematic review and previous ones[2,18,20,34,72,73,83,88,113,131] is its global approach to orofacial myofunctional reeducation (OMR), both active and passive, and its focus on the therapeutic strategies used by the various authors.

3. Materials and methods

The PRISMA[84] statement checklist (preferred evidence for systematic reviews and meta-analyses) was used as a guideline for conducting this systematic review. Figure 1 presents a flow chart of the search and selection process used.

3.1. The selection criteria

The following selection criteria were used.

3.1.1. Inclusion criteria

- Population of interest

The systematic review was limited to participants who met the following criteria: (1) polysomnographic diagnosis of OSA, (2) clinical symptoms of OSA, and (3) no syndromes and no other comorbid conditions such as, for example, head or neck injury, stroke, cancer or neurological disease.

- Types of studies and types of interventions

Only human clinical studies, written in English or French, published in peer-reviewed journals, which analysed the effects of active or passive orofacial myofunctional reeducation (or oropharyngeal exer-

cises), used alone or in combination with another treatment such as CPAP, in patients with OSA were included.

Studies were required to provide polysomnographic data, at least for apnoea and hypopnoea index (AHI), before and after treatment with OMR.

Randomised clinical trials comparing OMR with a placebo intervention or a controlled intervention and prospective cohort studies were included. Due to the paucity of relevant publications, retrospective cohort studies were also sought.

3.1.2. Judging criteria

The primary endpoint of this systematic review was a decrease in the apnoea/hypopnoea index (AHI) of at least five episodes per hour from baseline.

Secondary endpoints were an improvement in subjective sleep quality, sleep quality measured by nocturnal polysomnography and subjectively measured quality of life. The benefits and possible adverse effects of the strategies used and how they may affect the functionality of the upper airways were also recorded.

3.2. Information sources, search strategies and identification of studies

A systematic review of the literature was undertaken using the three electronic databases *Medline (via PubMed), Cochrane Library, Web of Science Core Collection*, in order to identify studies evaluating the efficacy of active or passive OMR in the treatment of OSAS in children, adolescents and adults published up to 4 September 2019.

The search strategy and identification of relevant studies used for the electronic search was based on the combination of the keywords "OSA" OR "obstructive sleep apnea" OR "sleep" OR "sleep apnea syndrome" AND "myofascial rééducation" OR "myofunctional therapy" OR "orofacial myotherapy" OR "oral myotherapy" OR "tongue exercises" OR "oropharyngeal exer- cises" OR "speech therapy" OR "upper airway exercises" OR "breathing exercises" OR "upper airway remodeling".

The eligibility of articles was determined in two phases. In the first phase, the two authors independently searched the three electronic databases and screened for studies that evaluated the effectiveness of OMR in the treatment of OSAS in children, adolescents and adults. If study abstracts were not available or were not self-explanatory, the full texts were retrieved and reviewed before a final decision was made. Once potentially eligible studies were selected, the full papers were obtained for the second phase of the selection process.

In this second stage, the same reviewers independently assessed the selected studies.

Articles present in several databases were only considered once.

Both authors searched the bibliographies of the selected articles for other relevant articles and grey literature was partially covered by consulting *Google Scholar*.

Papers that did not meet all the eligibility criteria were excluded. Disagreements between the two reviewers were discussed until a consensus was reached.

Where data were missing, additional information was requested from the authors of the studies by email.

3.3. Data collection

Data collection was done by both reviewers on standardised tables, and they then reviewed the extracted information.

The authors collected the following data: name of authors, year of publication, type of study, risk of study bias, number of subjects, demographic characteristics (gender, age), severity of OSAS, methodological parameters of OMR treatments and description of outcomes after OMR.

The data collected on the methodological parameters of OMR treatments were: therapeutic parameters (procedures, appointment frequency, treatment duration and therapeutic follow-up), assessment and reassessment parameters, physiological parameters (anthropometric measurements such as body mass index (BMI) and neck circumference before and after OMR, polysomnographic data such as apnoea-hypopnoea index (AHI) and minimum oxygen saturation (SpO2) before and after OMR), symptomatology (quality of life and sleep quality data, daytime sleepiness (*Epworth Sleepiness Scale*)[69] and snoring intensity and frequency).

Any inaccuracies or disagreements were resolved by re-examining the original document. If necessary, the authors of the selected studies were contacted and questioned about missing, inaccurate or incomplete data.

3.4. Risk of bias in individual studies

Both authors independently assessed the included studies using the twelve *Cochrane Back Review Group* (CBRG)[46] criteria for systematic reviews. These criteria, presented in Table 1 are combined with instructions[46] adapted from Van Tulder[132], Boutron, *et al.*[15] and the *Cochrane Handbook of Reviews of Interventions*[60].

Each criterion should be given one of three ratings: "yes", "no" or "uncertain".

A "yes" rating indicates that the criterion has been met and therefore suggests a low risk of bias.

Studies are assessed as being at "low risk of bias" when at least 6 of the 12 *Cochrane Back Review Group* (CBRG)[46] criteria have been met and the study does not have significant biases, such as a rate of lost to follow-up of more than 20% in one of the study groups. Studies with serious flaws, or those in which fewer than six of the twelve criteria are met, are assessed as having a "high risk of bias".

Any discrepancies that arose during the assessment of the quality of the studies were resolved by discussion between the reviewers.

4. Results

4.1. Selection of studies

A total of 1242 articles (*Medline*: 238; *Web of Science Core Collection*: 86; *Cochrane Library*: 50; *Google Scholar*: 868) were initially identified with electronic searches. After analysing the available titles and abstracts and removing duplicates, 23 articles were selected.

One article was selected after reviewing the bibliographies. A total of 24 articles were considered eligible and warranted full reading.

After reading the 24 articles in full, 14 were excluded on the basis of the selection criteria: one study of two cases[11] and 13 studies without a control gr oup[10,27,29,59,76,77,78,82,104,116,117,133,142].

In the end, 10 articles[28,35,36,53,56,63,65,85,125,134] meeting all the inclusion criteria were selected for this systematic review. The different stages of the selection process are described in the flow diagram (Fig. 1).

4.2. Description of the included studies

Six of the ten included studies focused on adult OSAS (Diaféria, *et al.*[35,36], Guimaraes, *et al.*[56], Ieto, *et al.*[65], Neumannova, *et al.*[85], Torres-Castro, *et al.*[125]) and four on paediatric OSAS (Chuang, *et al.*[28], Guilleminault, *et al.*[53], Huang, *et al.*[63], Villa, *et al.*[134]).

Table 1. Sources of risk of bias[46].

A	1. Was the method of randomisation adequate?	Yes/No/Uncertain
B	2. Has the treatment allowance been concealed?	Yes/No/Uncertain
C	Has knowledge of the interventions allocated been sufficiently avoided during the study?	
	3. Were the patients blind to the intervention?	Yes/No/Uncertain
	4. Were the carers blind to the intervention?	Yes/No/Uncertain
	5. Was the outcome assessor blind to the intervention?	Yes/No/Uncertain
D	Have incomplete outcome data been adequately addressed?	
	6. Was the lost to follow-up rate described and acceptable?	Yes/No/Uncertain
	7. Were all randomised participants analysed in the group to which they were assigned?	Yes/No/Uncertain
E	8. Were all results for the pre-determined outcome measures reported?	Yes/No/Uncertain
F	Other potential sources of bias:	
	9. Were the groups similar at baseline in terms of key prognostic factors?	Yes/No/Uncertain
	10. Were co-interventions avoided or similar?	Yes/No/Uncertain
	11. Was compliance acceptable in all groups?	Yes/No/Uncertain
	12. Was the timing of the outcome assessment the same in all groups?	Yes/No/Uncertain

The description of the studies, including the main characteristics of the sample, study objectives, type of sleep-disordered breathing, level of severity of OSA, intervention parameters and outcome measures, is presented in Table 2.

Of the ten studies[28,35,36,53,56,63,65,85,125,134] included in this systematic review, eight were randomised controlled trials[35,36,56,63,65,85,125,134], one was a prospective cohort study[28] and one was a retrospective cohort study[53].

Two randomised controlled trials (RCTs)[63,134] and a retrospective cohort study[53] have investigated the effectiveness of OMR as a means of reducing residual OSA in children after adenotonsillectomy.

The two RCTs by Diaferia, *et al.*[35,36] originally correspond to the same study and, although they each provide additional information, the data come from the same subjects, which explains the similarity of the results presented for these two studies in Results Tables 5 and 6.

All eight randomised controlled trials[35,36,56,63,65,85,125,134] included a control group, as expected for this type of study.

In seven of these RCTs, patients in the control group received sham OMR to minimise performance bias, with nasal lavage and deep breathing exercises[56], head movements without therapeutic function[35,36], nasal lavage alone[134], nasal lavage combined with deep breathing exercises and nocturnal nasal dilator strips during sleep[65], daily one-hour walk[85], daily half-hour walk combined with dietary and sleep recommendations[125].

For the RCT by Huang, *et al.*[63], the objective of the study was to compare the effects of active and passive OMR with the Michèle Hervy-Auboiron mandibular advancement device with tongue bead[28,63,79] set at 50% of maximum propulsion, and the RCT included two groups (active and passive OMR).

Two RCTs[35,36] that compared the effects of OMR alone or combined with CPAP included four groups of patients (OMR, CPAP, OMR + CPAP, control). Another RCT[85] also analysed the combination of OMR + CPAP with two groups (OMR + CPAP, control with CPAP alone).

Patients in the control group of the prospective cohort study[28] did not receive any treatment.

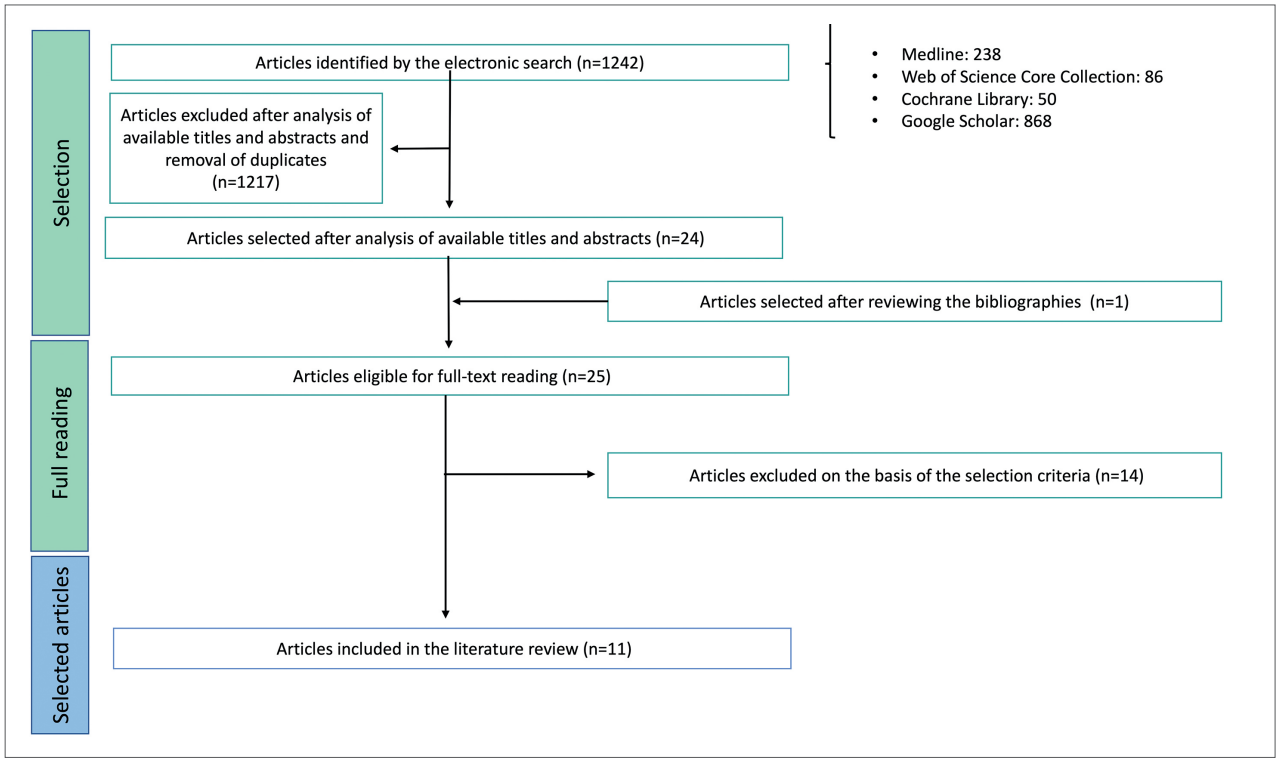

Figure 1
Flow diagram.

Table 2. Description of study data on subject characteristics, type of disorder, degree of OSA severity and treatment parameters.

Studies	Type of study	Characteristics of the sample	Objectives	Type and severity of OSAS	OMR treatment procedure (duration, frequency, exercises)	Judging criteria
Guimaraes, *et al.* 2009[56]	RCT	*Experimental group* N: 16 Age: 51.5 ± 6.8 Male: 63% AHI: 22.4 ± 4.8 BMI: 29.6 ± 3.8 *Control group* N: 15 Age: 47.7 ± 9.8 Male: 73% AHI: 22.4 ± 5.4 BMI: 31.0 ± 2.8	To assess, in patients with moderate OSAS, the effects of oropharyngeal exercises on the objective measurement of OSAS severity with polysomnography as well as on subjective sleep symptoms including snoring, daytime sleepiness and sleep quality	Moderate OSAS	Three months for all groups, with weekly visits *Experimental group* Nasal washing and oropharyngeal exercises Exercises of the soft palate, tongue and facial muscles Exercises in ventilation, phonation, swallowing and alternate chewing 1 x 30 min supervised session/week 1 session of 30 minutes at home/day *Control group* Nasal cleansing 3 times a day Deep breathing exercises through the nose in a sitting position 1 session of 30 mn supervised/week 1 x 30 min. session at home/day	*Main criterion* PSG AHI *Secondary criteria* Minimum arterial oxygen saturation (SaO2) Berlin questionnaire (snoring frequency) Epworth sleepiness scale [ESS] (subjective daytime sleepiness) Pittsburgh Sleep Quality Index
Diaféria, *et al.* 2013[35]	RCT	*Speech Therapy Only Group* N: 27 Age: 45.2 ± 13.0 Male: 100% AHI: 28.0 ± 22.7 BMI: 25.0 ± 7.4 ESS: 13.7 ± 3,2	To evaluate the effect of speech therapy, alone or combined with CPAP, on the quality of life of patients with OSAS	Mild, moderate and severe OSA	Three months for all groups *Speech Therapy Only Group* Oropharyngeal exercises 3 sessions of 20 mn at home/day *CPAP Group* Device with nasal mask, without humidifier and set to the optimal pressure according to the PSG of each patient	All outcomes were measured before and after treatment and after a 3-month washout period

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Studies	Type of study	Characteristics of the sample	Objectives	Type and severity of OSAS	OMR treatment procedure (duration, frequency, exercises)	Judging criteria
		CPAP Group N: 27 Age: 46.4 ± 9.1 Male: 100% AHI: 34.4 ± 22.4 BMI: 28.7 ± 3.3 ESS: 12.0 ± 2.1 *Speech Therapy and CPAP Group* N: 22 Age: 47.5 ± 10.9 Male: 100% AHI: 30.4 ± 19.8 BMI: 27.9 ± 2.4 ESS: 12.0 ± 2.6 *Control group* N: 24 Age: 42.9 ± 10.5 Male: 100% AHI: 27.8 ± 20.3 BMI: 28.6 ± 4.0 ESS: 12.8 ± 3.1			*Speech Therapy and CPAP Group* Combining the two protocols *Control group* Head movements without therapeutic function -3 sessions of 20 mn at home/day	PSG AHI Minimum arterial oxygen saturation (SaO2) Epworth sleepiness scale [ESS] Functional Outcome in Sleep questionnaire (FOSQ) WHOQOL-Bref questionnaire SF-36 Health questionnaire
Guilleminault, *et al.* 2013[53]	Retrospective cohort	*Group with myofacial reeducation* N: 11 Age: 7.3 ± 1.5 Boys/Girls: 23/1 AHI: 0.4 ± 0.3 Minimum SaO2 (%): 95 ± 1 Flow limitation (% total sleep time): 10 ± 10	To evaluate the impact of myofunctional rééducation (MR) in children with sleep disordered breathing referred for adenotonsillectomy, orthodontics, and myofunctional treatment in three different geographical areas	Mild, moderate and severe OSA	*Group with myofacial reeducation* Duration of 24 months Tongue and orofacial muscle strengthening exercises Regular supervised sessions as needed Several sessions at home/day	PSG at diagnosis, following adenotonsillectomy + orthodontics, and at long-term follow-up

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Studies	Type of study	Characteristics of the sample	Objectives	Type and severity of OSAS	OMR treatment procedure (duration, frequency, exercises)	Judging criteria
		Control group N: 13 Age: 7.3 ± 1.5 Boys/Girls: 23/1 AHI: 0.4 ± 0.3 Minimum SaO2 (%): 95 ± 1 Flow limitation (% total sleep time): 10 ± 10			*Control group* Lack of myofacial reeducation due to non-adherence to recommendations or abandonment	AHI Minimum SaO2 Flow limitation Myofunctional assessment Modified Mallampati score
Villa, *et al.* 2015[134]	RCT	*Experimental group* N: 14 Age: 6.01 ± 1.55 AHI: 4.87 ± 2.96 BMI (percentile): 81.85 ± 29.94 *Control group* N: 13 Age: 5.76 ± 0.82 AHI: 4.56 ± 3.22 BMI (percentile): 68.22 ± 28.68	To evaluate the effectiveness of oropharyngeal exercises as a means of reducing residual OSA in children after adenoamygdalactomy (AA)	Residual OSAS after AA	Two months, three meetings with the therapist *Experimental group* Nasal cleansing and oropharyngeal exercises 3 sessions at home/day, with 10-20 repetitions each time *Control group* Nasal cleansing 2 sessions at home/day, morning and evening	PSG before AA, 6 months after AA and after 2 months of exercise Improvement in OSA was defined as Δ AHI : (AHI at T1-AHI at T2)/ AHI at T1×100 Sleep Clinical Record (SCR) questionnaire Morphofunctional assessment
Ieto, *et al.* 2015[65]	RCT	*Experimental group* N: 19 Age: 48 ± 14 Male: 57.9% AHI: 15.6 ± 9.3 BMI: 28.3 ± 2.7	To assess the effects of oropharyngeal exercises on snoring in patients with minimal snoring symptoms and a diagnosis of primary snoring or mild to moderate OSA	Mild and moderate OSA	Three months for all groups, with weekly visits *Experimental group* Nasal cleansing Oropharyngeal exercises 1 supervised session/week 3 sessions of 8 mn at home/day	Objective snoring index and total snoring index obtained after recording snoring during PSG

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Studies	Type of study	Characteristics of the sample	Objectives	Type and severity of OSAS	OMR treatment procedure (duration, frequency, exercises)	Judging criteria
		Control group N: 20 Age: 45 ± 13 Male: 55% AHI: 15.1 ± 9.5 BMI: 28.3 ± 2.5			*Control group* Nasal cleansing Deep breathing exercises Nasal dilation strips during sleep 1 supervised session/week 3 sessions of 8 mn at home/day	AHI Epworth sleepiness scale [ESS] (subjective daytime sleepiness) Pittsburgh Sleep Quality Index Anthropometric assessment
Diaféria, *et al.* 2017[36]	RCT	*Myofunctional Therapy (MFT) group alone* N: 27 Age: 45.2 ± 13.0 Male: 100% AHI: 28.0 ± 22.7 BMI: 25.0 ± 7.4 ESS: 13.7 ± 3.2 *CPAP Group* N: 27 Age: 46.4 ± 9.1 Male: 100% AHI: 34.4 ± 22.4 BMI: 28.7 ± 3.3 ESS: 12.0 ± 2.1 *MFT + CPAP Group* N: 22 Age: 47.5 ± 10.9 Male: 100% AHI: 30.4 ± 19.8 BMI: 27.9 ± 2.4 ESS: 12.0 ± 2.6 *Control group* N: 24 Age: 42.9 ± 10.5 Male: 100% AHI: 27.8 ± 20.3 BMI: 28.6 ± 4.0 ESS: 12.8 ± 3.1	Assessing the effect of myofunctional treatment on CPAP compliance	Mild, moderate and severe OSA	Three months for all groups, with weekly visits for the placebo, MFT and MFT + CPAP groups Three visits for the CPAP group *MFT Group* Oropharyngeal exercises 3 sessions of 20 mn at home/day *CPAP Group* Device with nasal mask, without humidifier and set to the optimal pressure according to the PSG of each patient *MFT + CPAP Group* Combining the two protocols *Control Group* Head movements without therapeutic function 3 sessions of 20 mn at home/day	All outcomes were measured before and after treatment and after a 3-week washout period Assessment of compliance with CPAP PSG AHI Epworth sleepiness scale [ESS] Subjective assessment of snoring intensity and frequency Myofunctional assessment Modified Mallampati score

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Studies	Type of study	Characteristics of the sample	Objectives	Type and severity of OSAS	OMR treatment procedure (duration, frequency, exercises)	Judging criteria
Huang, *et al.* 2018[63]	RCT	*A-MFT Group (Active MFT)* N: 54 Age: 7.02 ± 2.44 Boys: 50% BMI: 15.61 ± 1.74 AHI: 2.47 ± 1.31 *P-MFT Group (Passive MFT)* N: 56 Age: 7.97 ± 3.08 Boys: 64.6% BMI: 17.04 ± 3.05 AHI: 6.00 ± 7.23	To evaluate the impact of active (A-MFT) or passive (P-MFT) myofunctional therapy in children with residual OSA after adenoamygdalectomy (AA) and children with OSA without indication for AA	Moderate and severe OSAS	12 months for all groups *A-MFT Group (Active MFT)* Isometric and isotonic exercises of the soft palate, tongue and facial muscles Exercises in ventilation, phonation, swallowing and alternate mastication 1 session of 20 mn at home/day Regular supervised sessions *P-MFT Group (Passive MFT)* Nightly wearing of the mandibular advancement device with tongue bead set at 50% of maximum mandibular propulsion Monthly check-up by the orthodontist	Clinical and PSG assessment at baseline, 3 months, 6 months and 12 months AHI Cephalometric assessment at baseline, 6 and 12 months
Neumannova, *et al.* 2018[85]	RCT	*Experimental group (CPAP +PR)* N: 15 Age: 53.87 ± 7.79 Neck circumference: 44.9 ± 10.3 BMI: 40.3 ± 9.4 AHI: 54.2 ± 27.4 *Control group (CPAP)* N: 20 Age: 54.05 ± 5.53 Neck circumference: 43.5 ± 4.6 BMI: 36.4 ± 5.4 AHI: 55.4 ± 28.9	To compare the short-term effects of CPAP combined with a reeducation programme, combining pulmonary rehabilitation, oropharyngeal and facial exercises, with CPAP, on OSA severity, ventilation and changes in body characteristics of a newly diagnosed patient.	Moderate and severe OSAS	6 weeks for all groups *Experimental group (CPAP +PR)* Individual aerobic training combined with therapeutic education, breathing exercises and respiratory muscle training 2 sessions of 60 mn/week with a personal physiotherapist Oropharyngeal exercises: soft palate, tongue and facial muscle exercises *Control group (CPAP)* A minimum of 60 mn of walking per day	All outcomes were measured before and after treatment *Main criterion* PSG AHI Oxygen Desaturation Index Epworth Sleepiness Scale Mean nocturnal SpO2 *Secondary criteria* Percentage of total sleep time with oxygen saturation below 90%. BMI Vital capacity and forced expiratory volume in 1 s Neck, waist and hip circumference

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Studies	Type of study	Characteristics of the sample	Objectives	Type and severity of OSAS	OMR treatment procedure (duration, frequency, exercises)	Judging criteria
Torres-Castro, *et al.* 2019[125]	RCT	*Experimental group* N: 14 Age: 64.5 (51.8–74) Male: 53.8% AHI: 30.5 (22.5–41.3) BMI: 31.3 (27.5–35) Neck circumference: 40 (37–41.5) *Control group* N: 13 Age: 67 (53–74.5) Male: 57.1% AHI: 37 (25.5–43.5) BMI: 27.1 (25.1–35.9) Neck circumference: 38 (37–41.5)	To evaluate the effects of a combined physical and oropharyngeal exercise programme on the apnoea-hypopnoea index in patients with moderate to severe	Moderate and severe OSAS	8 weeks for all groups *Experimental group* Brisk walking in urban areas for 30 mn, 3 times a week, usually supervised by a physiotherapist Oropharyngeal exercises Nutrition and sleep recommendations *Control group* Recommendations for diet and sleep Brisk walk for 30 minutes, at least 3 times a week	All outcomes were measured before and after treatment PSG or NRP AHI Oxygen Desaturation Index Epworth Sleepiness Scale Quebec Sleep Questionnaire Hospital Anxiety and Depression Scale (HADS) BMI Neck, waist and hip circumference
Chuang, *et al.* 2019[28]	Prospective cohort	*P-MFT Group (Passive MFT)* N: 17 Age: 7.95 ± 3.27 Boys: 77.5% BMI: 17.60 ± 3.72 AHI: 3.09 ± 2.56 *Control group* N: 40 Age: 7.65 ± 2.70 Boys: 76.5% BMI: 19.25 ± 3.98 AHI: 3.09 ± 2.56	To evaluate the effects of one year of passive myofunctional therapy (P-MFT) on craniofacial and airway morphology and quality of life in children with OSA	Moderate OSAS	12 months for all groups *Experimental group (Passive MFT)* Nightly wearing of the mandibular advancement device with tongue bead set at 50% of maximum mandibular propulsion Checked every 3 months by the orthodontist *Control group* No treatment	All outcomes were measured before and after treatment PSG AHI Lateral cephalometric assessment Quality of life questionnaire (OSA-18)

Abbreviations: RCT, randomised controlled trial; OSAS, obstructive sleep apnoea and hypopnoea syndrome; BMI, body mass index; PSG, polysomnography; NRP, nocturnal respiratory polygraphy; AHI, apnoea-hypopnoea index; CPAP, continuous positive airway pressure; PR, pulmonary rehabilitation, oropharyngeal and facial exercises program.

The control group in the retrospective cohort study[53] consisted of patients who did not complete the myofacial reeducation program due to non-compliance or drop-out.

Active, non-device based OMR procedures differed between studies in the choice of exercises, number of repetitions, frequency and duration of daily practice (from six weeks to one year), while maintaining a common approach. The authors mainly used a series of "oropharyngeal" exercises, derived from speech therapy and physical therapy. Isometric and isotonic exercises were performed to optimise muscle tone and mobility, to adjust the position of the soft tissues (soft palate, pharyngeal constrictor muscles, supra-hyoid muscles, tongue, cheeks and lips) and to improve the orofacial functions of ventilation, mastication, swallowing and phonation. The authors did not provide any justification for their choice of the type of exercises, frequency and duration of rehabilitation sessions. Table 3 shows an example of oropharyngeal exercises used in the Neumannova RCT[85].

The potential for the effects of OMR to persist remains an issue, particularly for orthodontists. Take the example of lip tone exercises, which are often prescribed to help the patient regain lip competence. One study showed that lip training with an oral screen for nine months did increase lip strength, but that it then decreased as measured ten months after the muscle training stopped[123]. It may therefore seem appropriate to extend the OMR therapy programs to maintain the results obtained previously

Table 3. Description of oropharyngeal exercises used in the Neumannova RCT[85].

Type of exercises	Description of the exercises
Tongue exercises	- Push anterior half of the tongue against the hard palate for 5 s., keep the jaw open throughout the exercise, relax the tongue for 8 s., 10 repetitions, three times a day. - Open the mouth widely, try to touch the chin with the tip of the tongue, hold this position for 5 s., place the tongue into the mouth and relax for 8 s., 10 repetitions, three times a day.
Soft palate exercises	- Pronounce an oral vowel “A, E, I, O, U” intermittently (isotonic exercise) and continuously (isometric exercise), five repetitions, once a day. - Breathe in through the nose, breathe out through the mouth, during breathe out period press the lips together, maintain the blowing for 5 s., five repetitions, three times a day.
Exercises for the cheeks, throat, and neck	- Tilt the head back, stick the tongue out and upward (“try to touch the ceiling with the tip of the tongue”), hold it for 5 s., then move the head into an upright position and relax the tongue in the mouth for 8 s., 10 repetitions, once a day. - Tilt the head back, gently bite the tongue and try to swallow once, then move the head in an upright position and relax the tongue in the mouth for 8 s., five repetitions, once a day. - Place the index finger inside the cheek, place the thumb on outside the cheek, pull the cheek outward with the fingers, at the same time contract the cheek muscle to resist the pulling for 5 s., relax for 8 s., 10 repetitions, once a day.
Exercises for lips and jaw	- Purse the lips, hold the position for 10 s., relax for 12 s., five repetitions, once a day. - Purse the lips with the mouth wide open, hold the position for 5 s., relax for 8 s., five repetitions, once a day. - Place a hand under the chin, attempt to open the mouth for 5 s., but with the hand pushing against the lower jaw (the task is to stop the mouth opening), relax for 8 s., 10 repetitions, once a day.

* Oral vowels are pronounced with the soft palate raised, which closes off the passage of air through the nose. Nasal vowels are pronounced with the soft palate lowered, allowing air to pass through the mouth and nose.

Table 4. Methodological quality of studies assessed according to the Cochrane Back Review Group[46] risk of bias criteria.

Type of exercises	Methodological quality according to the CBRG[46] risk of bias criteria												Total	Quality	Conflit of interest
	1	2	3	4	5	6	7	8	9	10	11	12			
Guimaraes, *et al.* 2009[56]	NS	N	Y	N	Y	Y	Y	Y	Y	Y	Y	Y	9	High	No
Diaféria, *et al.* 2013[35]	NS	N	Y	N	NS	N	Y	Y	Y	Y	Y	Y	7	High	No
Guilleminault, *et al.* 2013[53]	NS	N	N	N	Y	Y	Y	Y	Y	Y	Y	Y	8	High	No
Villa, *et al.* 2015[134]	NS	N	Y	N	Y	Y	Y	Y	Y	Y	Y	Y	9	High	No
Ieto, *et al.* 2015[65]	NS	N	Y	N	Y	Y	Y	Y	Y	Y	Y	Y	9	High	No
Diaféria, *et al.* 2017[36]	NS	N	Y	N	Y	N	Y	Y	Y	Y	Y	Y	8	High	No
Huang, *et al.* 2018[63]	NS	N	N	N	Y	N	Y	Y	Y	Y	N	Y	6	High	No
Neumannova, *et al.* 2018[85]	NS	N	Y	N	N	Y	Y	Y	Y	Y	Y	Y	8	High	No
Torres-Castro, *et al.* 2019[125]	Y	Y	Y	N	Y	Y	Y	Y	Y	Y	Y	Y	11	High	No
Chuang, *et al.* 2019[28]	NS	N	N	N	Y	Y	Y	Y	Y	Y	Y	Y	8	High	No

Y: yes, N: no, NS: not specified by the authors of the study.

and to combine therapeutic education with OMR[3].

This issue is even more important for OMR treatments for OSAS, which is a particularly restrictive approach and requires unfailing patient compliance, which explains the many dropouts. The only studies that have raised this important issue of compliance are a retrospective cohort study[53], with assessment between 22 and 50 months after completion of OMR, and four RCTs[28,35,36,63]. Two of the RCTs addressed this issue with medium- and long-term follow-up[28,63] and two repeated the measures after a 3-week washout period[35,36].

The data collected in the ten studies in this systematic review were physiological or symptomatological.

The main physiological data were either obtained by polysomnography, for apnoea and hypopnoea index per hour of sleep (AHI) and minimum blood oxygen saturation (minimum SpO2), or were anthropometric measurements, including body mass index (BMI) and neck circumference.

The main symptomatological data included measures of quality of life, including the *Whoqol-Bref, Functional Outcome in Sleep Questionnaire* (FOSQ) and the SF-36 health questionnaire, and of sleep quality, using the Pittsburgh Sleep Quality Index. Subjective daytime sleepiness with the Epworth Sleepiness Scale (ESS) and snoring intensity and frequency with a visual analogue scale, the Berlin Questionnaire and snoring recording during polysomnography were also assessed.

4.3. Methodological quality of the included study

All included studies met at least six of the *Cochrane Back Review Group (CBRG)*[46] criteria (Tab. 1) and were not subject to significant bias, such as too many patients being lost to follow-up in one of the study groups, so they were all assessed as being at "low risk of bias" (Tab. 4).

Table 5. Results of orofacial myofunctional reeducation (OMR) in terms of physiological data.

Studies	Anthropometric measurements				Polysomnography			
	BMI (kg/m 2)		NC (cm)		AHI (nb/hour)		SpO2 minimum (%)	
	Before	After	Before	After	Before	After	Before	After
Guimaraes, *et al.* 2009[56]	29.6 ± 3.8	29.5 ± 4.3	39.6 ± 3.6	38.5 ± 4.0* (P=0.01)	22.4 ± 4.8	13.7 ± 8.5* (P<0.01)	83 ± 6	85 ± 7*
Diaféria, *et al.* 2013[35] ∏	25.0 ± 7.4	26.7 ± 2.9	41.6 ± 3.7	41.5 ± 2.3	28 ± 2.7	13.9 ± 18.5* (P<0.001)	83.7 ± 7.7	84.9 ± 8.8
Guilleminault, *et al.* 2013[53]	NR	NR	NR	NR	5.3 ± 0.3 (without OMR)	0.5 ± 0.4* (with OMR) (P=0.001)	91 ± 1.8	96 ± 1* x2 test (P=0.1)
Villa, *et al.* 2015[134]	81.85 ± 29.94	NR	NR	NR	4.87 ± 2.96	1.84* (P=0.004)	NR	NR
Ieto, *et al.* 2015[65]	28.1 ± 2.7	28.2 ± 2.8	37.9 ± 2.5	37.5 ± 2.4** (P<0.05)	25.4 (22.1-28.7) ֍	18.1 (15.4-24.1) ֍*** (P=0.17)	85.5 ± 7.5	83.8 ± 8.9
Diaféria, *et al.* 2017[36] ∏	25.0 ± 7.4	26.7 ± 2.9	41.6 ± 3.7	41.5 ± 2.3	28 ± 22.7	13.9 ± 18.5* (P<0.001)	83.7 ± 7.7	84.9 ± 8.8
Huang, *et al.* 2018[63] Ω	17.04 ± 3.05	18.53 ± 3.99	NR	NR	6.00 ± 7.23	2.44 ± 2.28* (P=0.001)	NR	NR
Neumannova, *et al.* 2018[85]	40.3 ± 9.4	39.6 ± 9.1* (P<0.01)	44.9 ± 10.3	43.7 ± 3.5* (P<0.005)	54.2 ± 27.4	4.3 ± 3.9* (P<0.0001)	NR	NR
Torres-Castro, *et al.* 2019[125]	31.3 (27.5-35) ֍	30.2 (27.3-34.7)* (P=0.003) ֍	40(37-41.5) ֍	38.8(37.4-40.5) ֍	30.5(22.5-41.3) ֍	34.5(14.5-45) ֍	NR	NR
Chuang, *et al.* 2019[28]	17.6 ± 3.72	NR	NR	NR	3.75 ± 2.48	2.16 ± 1.80* (P=0.002)	SaO2 minimum 90.13 ± 4.01	SaO2 minimum 91.95 ± 3.56* (P=0.001)

Data are presented with their mean and standard deviation, except for ֍ presented with the median, minimum and maximum.
Ω: only the results of the group with passive myofunctional therapy (P-MFT) were presented, as no patients in the active myofunctional therapy (A-MFT) group completed the study.
∏: only results from the OMR group are presented.
Abbreviations: OMR, orofacial myofunctional reeducation; NR, no record of this data, which did not correspond to the objective of the study or which the study authors did not retain; BMI, body mass index; NC, neck circumference; AHI, apnoea and hypopnoea index per hour of sleep; SpO2 minimum, minimum blood oxygen saturation; CPAP, continuous positive airway pressure; *, P statistically significant (<0.05) T-test; **, P statistically significant for comparisons using repeated measures analysis of variance; ***, P variation only for the moderate OSAS group.

Table 6. Results of orofacial myofunctional reeducation (OMR) in terms of symptomatological data.

Studies	Symptomatology							
	Quality of life Quality of sleep		ESS		Intensity of snoring		Frequency of snoring	
	Before	After	Before	After	Before	After	Before	After
Guimaraes, *et al.* 2009[56]	Pittsburgh 10.2 ± 3.7	Pittsburgh 6.9 ± 2.5*	14 ± 5	8 ± 6* (P=0.01)	3 (3-4) ֍	1 (1-2) * ֍ (P=0.001)	4 (4-4) ֍	3 (1.5 - 3.5) * ֍ (P=0.001)
Diaféria, *et al.* 2013[35] ∏	FOSQ WHOQOL-Bref SF-36	FOSQ WHOQOL-Bref* (P<0.001) SF-36	13.7 ± 3.2	7.5 ± 3.7* (P<0.001)	8.5 ± 2.3	4.9 ± 3.2* (P<0.001)	7.7 ± 2.3	4.3 ± 2.8* (P<0.001)
Guilleminault, *et al.* 2013[53]	NR	NR	NR	NR	NR	NR	NR	NR
Villa, *et al.* 2015[134]	SCR 4.07 ± 2.22	NR	NR	NR	NR	NR	NR	NR
Ieto, *et al.* 2015[65]	Pittsburgh 6 ± 3.2	Pittsburgh 4 ± 2.6* (P=0.04)	7 (3-11) ֍	7 (4-10) ֍	2 (2-3) S 4 (2.5-4) P ֍	2 (1-2) S 1 (1-2) P* ֍ (P=0.03)	3 (2-4) S 4 (3-4) P ֍	2 (1-4) S 2 (1.5-3) P* ֍ (P=0.04)
Diaféria, *et al.* 2017[36] ∏	NR	NR	13.7 ± 3.2	7.5 ± 3.7* (P<0.001)	8.5 ± 2.3	4.9 ± 3.2* (P<0.001)	7.7 ± 2.3	4.3 ± 2.8* (P<0.001)
Huang, *et al.* 2018[63] Ω	NR	NR	NR	NR	Snoring index (nb/h) 212.91	Snoring index (nb/h) 83.16* (P=0.046)	NR	NR
Neumannova, *et al.* 2018[85]	NR	NR	12.9 ± 4.7	5.7 ± 4.1 * (P<0.0005)	NR	NR	NR	NR
Torres-Castro, *et al.* 2019[125]	QSQ 180.5 (151-200) ֍	QSQ 186 (153.8-201) ֍	8 (3-13) ֍	8 (4-10.3) ֍	NR	NR	NR	NR
Chuang, *et al.* 2019[28]	OSA-18 55.85 ± 17.44	OSA-18 45.51 ± 14.44* (P=0.000)	NR	NR	Snoring index (nb/h) 212.91	Snoring index (nb/h) 212.91	NR	NR

Data are presented with their mean and standard deviation, except for ֍ presented with the median, minimum and maximum.
Ω: only the results of the group with passive myofunctional therapy (P-MFT) were presented, as no patients in the active myofunctional therapy (A-MFT) group completed the study.
∏: only the results of the group with OMR alone are presented.
Abbreviations: NR, no record of this data, which did not fit the study objective or was not retained by the study authors; ESS, Epworth Sleepiness Scale (subjective daytime sleepiness); *, P significant (<0.05) T-test; S, information given by the research subject; P, information given by the sleep partner; QSQ, Quebec Sleep Questionnaire; OSA-18, OSA-18 Quality of Life Questionnaire.

4.4. Results of the included studies

4.4.1. General presentation of the results

The main results of orofacial myofunctional rehabilitation (OMR) have been grouped in two tables, one where the results are described in terms of physiological data (Tab. 5), the other in terms of symptomatological data (Tab. 6).

4.4.2. Results obtained with active OMR

Active OMR is defined as OMR that is not associated with the use of a custom-made appliance[29,63] or prefabricated functional appliances[77,78,101,130,138,140] as is passive OMR.

The patient performs isotonic and isometric exercises that target the oral, oropharyngeal structures and are combined with specific ventilation, swallowing and chewing exercises. As dysfunction of the orofacial and pharyngeal muscles[33] and impaired oropharyngeal control can contribute to airway collapse[41], a contribution of active OMR to the management of OSA is sought.

4.4.2.1. Effects of active OMR on physiological variables

4.4.2.1.1. Effects of active OMR on PSG variables

Four[35,36,56,85] of the six RCTs dedicated to adults showed a significant reduction in AHI.

The three studies in children, two RCTs[63,134] and one prospective cohort study[7], all showed a significant decrease in AHI.

A statistically significant increase in the percentage of arterial minimum oxygen saturation was found in an adult RCT[56] and a prospective study in children[28].

4.4.2.1.2. Effects of active OMR on anthropometric variables

Measurements of neck circumference, abdominal circumference and BMI are anthropometric predictors of the severity of OSAS[96,124, 62]. Pharyngeal critical closure pressure is associated with obesity[50], decreased upper airway elasticity and is observed in obese patients with a large neck circumference[61,67].

The statistically significant decrease in neck circumference measured in the studies of Guimaraes, *et al.*[56], Ieto, *et al.*[65], Neumannova, *et al.*[85] suggests the possibility of upper airway remodelling by active OMR exercises. The decrease in neck circumference is correlated with a statistically significant reduction in AHI in the studies by Guimaraes, *et al.*[56] and Neumannova, *et al.*[85].

A statistically significant decrease in BMI is reported in the only study by Neumannova, *et al.*[85].

4.4.2.1.3. Effects of active OMR on orofacial myofunctional status

Assessment of myofunctional status should help to determine whether the effects of OMR in OSAS patients are related to improved muscle and orofacial function. Surprisingly, only the studies by Guilleminault, *et al.*[53], Villa, *et al.*[134] and Diaféria, *et al.*[36] included a myofunctional assessment. Interpretation and comparison of the results could not be conducted due to the lack of a standardised assessment tool.

In the retrospective cohort study by Guilleminault, *et al.*[53], myofunctional assessment of the orofacial region of the control group, who had not had myofacial reeducation due to non-compliance or drop-out, showed that the patients had an abnormally low tongue position in the mouth when awake. Twelve of the thirteen patients in the control group were unable to make tongue clicks, ten were unable to move their tongue upwards when asked to try to touch their nose with the tip of their tongue, four had difficulty holding a button between their lips, and one had difficulty swallowing when drinking rapidly. All subjects admitted to having a preferential side of unilateral chewing, and nine subjects had a slight asymmetry of the masseter muscles when assessing active contraction. Finally, all subjects were assessed with abnormal orofacial muscle tone when awake.

In contrast, all eleven patients in the OMR group were rated as normal on myofunctional assessment.

Diaféria, *et al.*[36] reported that the modified Mallampati score improved in the OMR and CPAP + OMR groups and was correlated with increased tongue and soft palate strength.

Villa, *et al.*[134] evaluated the effectiveness of oropharyngeal exercises as a means of reducing residual OSA in children after adenotonsillectomy. The exercises prescribed were of three types: (1) ventilatory rehabilitation, (2) lip closure and lip tone exercises, and (3) tongue posture exercises. Active OMR significantly increased the number of patients with nasal

ventilation, effective lip closure and good lip tone. No significant improvement was observed for patients in the control group.

4.4.2.2. Effects of active OMR on symptomatological variables

4.4.2.2.1. Effects of active OMR on quality of life and sleep quality

The studies by Diaféria, *et al.*[35,36] and Chuang, *et al.*28 showed a statistically significant improvement in quality of life. The RCTs of Diaféria, *et al.*[35,36] reported this improvement in quality of life for the OMR and OMR + CPAP groups, but not for the CPAP and control groups.

The studies by Guimaraes, *et al.*[56] and Ieto, *et al.*[65] showed a statistically significant improvement in sleep quality.

4.4.2.2.2. Effects of active OMR on daytime sleepiness

The Epworth Sleepiness Scale (ESS), proposed by Johns[69], is generally used to assess daytime sleepiness. The ESS assesses the propensity to sleep in eight different situations, with a total score ranging from zero (absence) to 24 (intense).

The RCT of Guimaraes, *et al.*[56] reported a statistically significant improvement in ESS at the end of treatment with OMR. The RCTs of Diaféria, *et al.*[35,36] also showed a significant improvement in ESS at the end of OMR treatment in the OMR, OMR + CPAP and CPAP groups. Measurement of ESS at the end of three weeks of therapeutic withdrawal after intervention showed that the improvement in SSE remained statistically significant in the OMR and CPAP groups.

4.4.2.2.3. Effects of active OMR on snoring intensity and frequency

Jacques Talmant, *et al.*[120] pointed out that structural changes, secondary to the vibratory trauma caused by snoring, can affect each component of the pharyngeal structures[43] and contribute to the collapsibility of this segment of the airways. They may increase the risk of carotid atherosclerosis[75] and bilateral carotid artery stenosis[32].

A statistically significant decrease in snoring intensity and frequency was observed in studies conducted by Guimaraes, *et al.*[56], Ieto, *et al.*[65] and Huang, *et al.*[63].

The studies by Diaféria, *et al.*[35,36] showed a significant decrease in snoring intensity and frequency for the OMR, OMR + CPAP and CPAP groups after treatment and in the OMR + CPAP and CPAP groups when compared to the control group.

At the end of three weeks of therapeutic withdrawal after intervention, the reduction in snoring intensity and frequency was statistically significantly maintained in the OMR group but did not persist in the OMR + CPAP and CPAP groups.

4.4.2.3. Contribution of active OMR to the treatment of OSA with continuous positive airway pressure (CPAP)

The objective of the RCT by Diaferia, *et al.*[35] was to assess the effect of OMR, either alone or in combination with CPAP, on the quality of life of patients with OSAS. Analysis of the quality of life data showed a statistically significant improvement in the physical health dimension (Whoql-Bref questionnaire) for the OMR and OMR + CPAP groups. The functional capacity dimension (SF-36 health questionnaire) improved statistically significantly in the OMR group. Quality of life, assessed with the FOSQ, did not change significantly in any of the four study groups.

The same team of Diaféria, *et al.* had evaluated the effect of OMR on adherence to CPAP in another RCT[36]. Although OMR did not reduce the level of CPAP required, it significantly increased adherence to CPAP. The mean adherence was 55% in the control group, 63% in the OMR group, 30% in the CPAP group and 65% in the combined CPAP + OMR group. Weekly therapy education and support during the sessions may have contributed to greater adherence to CPAP when combined with OMR (65%) than when prescribed alone (30%).

4.4.3. Results obtained with the passive OMR

To address the frequent lack of compliance observed in OMR programs, authors have proposed the use of passive OMR. This is named to reflect the assistance provided to the patient by wearing a custom-made appliance[29,63] or prefabricated functional appliances[77,78,101,130,138,140], the latter with the additional objective of modifying the shape and relationship of the dental arches. The inclusion of these appliances in the OMR procedures requires the practitioner to follow up rigorously, in order to continuously assess the potential adverse effects of the appliances on the dental arches, especially the vestibuloversion of the mandibular incisors.

The studies by Levrini[77,78] investigated the effect of a prefabricated myofunctional device for the treatment of mild to moderate paediatric obstructive sleep apnoea. They could not be included in the systematic review due to insufficient methodological quality, mainly related to the absence of a control group.

Studies by Huang, *et al.*[63] and Chuang, *et al.*[28] investigated the effects of passive OMR using the custom-made one-piece mandibular advancement orthosis designed by Michèle Hervy-Auboiron (patent number: EP 13753289.1; US14/420499). It is attached to the maxilla by stabilizing clasps placed between the maxillary second premolars and first molars. It has a freely rotating ball that is set at the anteroinferior end of the orthosis. The orthosis is constructed with moderate mandibular propulsion, at 50% of maximum mandibular advancement. The patient wears the orthosis only during sleep. The patient is asked to rotate the ball just before falling asleep in order to obtain lingual propulsion and upper airway clearance. The objective is to induce a persistent lingual propulsion response in case of apnea.

The study by Huang, *et al.*[63] aimed to assess the impact of active or passive OMR in children with residual OSA after adenotonsillectomy (AA) and in children with OSA without an indication for AA. It showed a statistically significant decrease in snoring intensity and AHI. Cephalometric assessment of the subjects showed no adverse effects from nighttime use of the orthosis set at 50% of maximum mandibular propulsion, and a significant increase in upper airway.

The study by Chuang, *et al.*[28] evaluated the effects of one year of passive OMR on the craniofacial and airway morphology and quality of life of children with OSA. She reported a statistically significant improvement in quality of life (OSA-18) and AHI during sleep and REM AHI. Cephalometric assessment of the patients showed significant improvement in linear mandibular growth (Co-Gn) and upper airway morphology.

5. Discussion

5.1. Summary of the main evidence

This systematic review, which includes ten studies on the effects of OMR on OSAS, six in adults (Ieto, *et al.*[65], Diaféria, *et al.*[35,36], Torres- Castro, *et al.*[125], Guimaraes, *et al.*[56], Neumannova, *et al.*[85]) and four in children (Chuang, *et al.*[28], Guilleminault, *et al.*[53], Huang, *et al.*[63], Villa, *et al.*[134]), leads to five main conclusions.

5.1.1. OMR allows for a reduction in AHI

In adults, OMR provides a statistically significant reduction in AHI from 28.74% (Ieto, *et al.*[65]) to 92.06% (Neumannova, *et al.*[85]), a study that reported a decrease in pre- and post-OMR AHI from 54.2 ± 27.4 to 4.3 ± 3.9* (P < 0.0001).

In children, OMR provides a statistically significant reduction in AHI of up to 90.6% in the retrospective cohort study by Guilleminault, *et al.*[53], in which the M ± SD was 0.5 ± 0.4* (P = 0.001) with OMR and 5.3 ± 0.3 without OMR. The RCT that reported the largest significant decrease was Villa, *et al.*[134], with a mean AHI that decreased by 62.22%, from 4.87 ± 2.96 before OMR to 1.84*, P = 0.004 after OMR.

The persistence of the effect of OMR on AHI is variable. The study by Diaféria, *et al.*[35] shows a decrease in AHI significance after three months of OMR, and a loss of significance after three weeks of therapeutic withdrawal. The study by Guilleminault, *et al.*[53] reported that eleven children remained cured of OSA (AHI of 0.5 ± 0.4/h) 22 to 50 months after completion of the OMR program.

5.1.2. OMR allows a reduction in the intensity and frequency of snoring

OMR can significantly reduce snoring, both subjectively and objectively (Guimaraes, *et al.*[56], Ieto, *et al.*[65], Huang, *et al.*[63], Diaféria, *et al.*[35,36]). The studies by Diaféria, *et al.*[35,36] reported a persistence of the effect of OMR with the maintenance of the decrease in snoring intensity and frequency in the OMR group at the end of the three weeks of therapeutic withdrawal after intervention.

5.1.3. OMR helps to reduce daytime sleepiness

OMR can reduce daytime sleepiness, as evidenced by statistically significant improvement in ESS at the end of OMR treatment (Guimaraes, *et al.*[56], Diaféria,

et al.[35,36]). The studies by Diaféria, *et al.*[35,36] showed that at the end of three weeks of therapeutic withdrawal after intervention the improvement in ESS remained statistically significant in the OMR and CPAP groups.

5.1.4. OMR improves adherence to CPAP treatment

The RCT by Diaféria, *et al.*[36] reported a significant increase in adherence to CPAP. The mean adherence was 55% in the control group, 63% in the OMR group, 30% in the CPAP group and 65% in the combined CPAP + OMR group.

5.1.5. Passive OMR is effective and increases compliance with reeducation

Passive OMR, with the assistance of a custom-made orthosis[29,63], increases compliance with reeducation (Huang, *et al.*[63]). None of the fifty-four patients in the active OMR group completed the one-year therapy follow-up, whereas 88.88% of the fifty-six patients in the passive OMR group completed the study[63].

Passive OMR resulted in a statistically significant decrease in snoring intensity, AHI and a significant increase in upper airway (Huang, *et al.*[63], Chuang, *et al.*[28]).

5.2. Limitations

A first limitation is the low compliance of adult and pediatric patients with their active OMR program. Another limitation is the difficulty of keeping participants enrolled in the studies and convincing them to undergo the complex end-of-study tests again. This makes it particularly difficult to assess the persistence of the effects of OMR.

Passive OMR appears to provide an effective response to this lack of compliance when based on the custom-made one-piece mandibular advancement orthosis designed by Michèle Hervy-Auboiron[29,63]. The other two studies (Levrini, *et al.*[77,78]), which used a prefabricated myofunctional appliance for the treatment of paediatric obstructive sleep apnoea, could not be included in the systematic review because, like all uncontrolled case series, their level of evidence was insufficient to lead to recommendations.

Continued attention should be paid to monitoring possible adverse effects of devices used for passive OMR on the dental arches, including vestibuloversion of the mandibular incisors.

The question arises as to the mode of action of the orthosis used in the studies by Huang, *et al.*[63] and Chuang, *et al.*[28]. Is its real efficiency only linked to a lingual propulsion and a clearing of the upper airways by the patient incited to rotate the ball located at the anteroinferior end of the orthosis, just before falling asleep? Is it also and partially related to the construction of the orthosis with a moderate mandibular propulsion, at 50% of the maximum mandibular advancement[7,9,141]?

Another limitation is the selection of studies. We included in this review randomised clinical studies comparing OMR with a placebo or controlled intervention and prospective cohort studies. Due to the paucity of relevant publications, we also searched for retrospective cohort studies and included the 2013 study by Guilleminault, *et al.*[53]. As with other retrospective cohort studies (case-control study), this study has a risk of selection bias[4].

The ten studies included in this systematic review had various methodological limitations, mainly related to the difficulty of recruiting compliant patients to participate in OMR treatment and keeping them in the study. For example, in the study by Chuang, *et al.*[28], the control group had milder forms of OSA than the treatment group, the sample size of the control group was significantly smaller than that of the treatment group, and the adenoids and tonsils of the control group were larger than those of the treatment group.

Other limitations relate to the short duration of OMR programs, for example six weeks in the study by Neumannova, *et al.*[85], which limits the validity of the results.

Also, due to the difficulty of recruiting participants, sample sizes are often small, which leads to low power of statistical tests.

Finally, OMR is based on an integrative approach to an exercise programme and therefore does not allow the specific effects of each exercise on the overall outcome to be determined.

6. Conclusions

Obstructive sleep apnoea syndrome (OSAS) is a widespread and under-diagnosed condition, making it a major public health and safety issue.

Orofacial myofunctional reeducation (OMR) has been shown to be effective in the multidisciplinary management of OSAS in children, adolescents and adults and is prescribed at several stages of management.

The ten studies included in this systematic review on the effects of OMR on OSAS show that OMR significantly reduces AHI by up to 90.6% in children and 92.06% in adults. It significantly reduces the intensity and frequency of snoring, contributes to a reduction in daytime sleepiness, limits the recurrence of OSA symptoms after adenotonsillectomy in children and improves adherence to CPAP therapy.

To address the frequent lack of compliance observed in OMR programs, some authors have proposed the use of passive OMR, with the assistance of a custom-made appliance or prefabricated functional appliances, the latter with the additional objective of modifying the shape and relationship of the dental arches.

The integration of these devices into OMR procedures requires the practitioner to carefully monitor the potential adverse effects of the appliances on the dental arches, particularly the vestibuloversion of the mandibular incisors.

Passive OMR, with the assistance of a custom-made ball orthesis, increases compliance with reeducation, allows a significant decrease in snoring intensity, AHI and a significant increase in the upper airway.

Prospective studies with large samples would be useful to better evaluate the persistence of the effects of OMR and the possibilities offered by passive OMR, with the ball mandibular advancement orthosis designed by Michèle Hervy-Auboiron or prefabricated functional appliances, to compensate for the frequent lack of compliance observed during active OMR treatments.

Links of interest

The authors declare that they have no interest in the data published in this article.

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