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Consensus statement on the orofacial myofunctional assessment and therapy in patients with OSA: proposal of an international Delphi Method process

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ABSTRACT

Orofacial myofunctional therapy has been shown to be effective in the multidisciplinary management of obstructive sleep apnoea syndrome in children, adolescents, and adults, and is prescribed at several stages of such management. However, given the lack of consistency in treatment protocols and outcome measures, the lack of research on the type, frequency and duration of myofunctional exercise and the specific phenotypes that may benefit from it, there is a need to define best practice guidelines.

A Delphi consensus guideline on orofacial myofunctional assessment and treatment for patients with OSA was initiated by Marc Richard Moeller. The first two stages of the Delphi consensus recommendations have been completed. The third stage, the development of the questionnaire, will take place at the joint meeting of the World Sleep Society and the Academy of Applied Myofunctional Sciences in March 2022 in Rome. The conclusions of the Delphi consensus recommendations will be published in the following year and widely disseminated.

KEYWORDS

Obstructive sleep apnea syndrome, orofacial myofunctional therapy, consensus statement, Delphi method process.

INTRODUCTION

Orofacial myofunctional therapy is the re-education of the muscles, functions, and rest postures of the orofacial complex, involving the lips, tongue, cheeks, mandible, sucking, chewing, swallowing, speech articulations and more. Myofunctional therapy has been used as a clinical solution for several decades with documented success, worldwide, addressing orofacial dysfunctions in patients of all ages and with a wide range of disorders and comorbidities.

Only recently there have been enough studies published on the application of myofunctional therapy, many with sleep disorders, reaching an increased level of medical consideration and a higher level of evidence such as meta-analyses and systematic reviews. Although the results of these higher level of evidence studies are sometimes inconclusive, conclusions have all suggested that the outcomes are powerful enough that myofunctional therapy both should be considered as a treatment and that further research should be conducted. Yet with a lack of homogeneity in treatment protocols and outcome measurements, as well as a dearth of research on type, frequency, and duration of myofunctional exercise and the specific phenotypes that may benefit, a great deal more research is needed. This paper proposes just that, and a process by which consensus may be developed around the next steps that should be undertaken by leaders and allied professionals concerned with the potential for advancement of this promising, emerging field.

Obstructive breathing disorders are characterized by the partial or complete reduction of the airflow during sleep. The resulting flow limitations, apnoeas, and hypopnoeas induce sleep fragmentation, intermittent hypoxia, and hypercapnia, as well as increased intra- thoracic pressure swings during the breathing effort. These breathing disorders during sleep range from snoring to obstructive sleep apnea (OSA)[34].

OSA is an endemic chronic disease that affects a high percentage of the pediatric and adult population, currently affecting an estimated one billion people[5]. Most patients with OSAS remain undiagnosed, making OSA a major public health and safety issue[34]. The prevalence of adult OSA has been reported to be as high as 30% in the Brazilian population aged from 18 to 80 years old[41], and a recent study in Switzerland confirmed moderate to severe OSA in 23% of the women and almost 50% of the men, when considered AHI>15 e/h[21].

Adult OSA is associated with an increased risk of health problems, including cardiovascular disease, carbohydrate-lipid metabolism disorders, and cancer. Besides these, there is an important link between chronic pain and sleep disorders, especially orofacial pain [5,34]. Pain is exacerbated by OSA, and pain disturbs sleep. Most orofacial pain disorders include orofacial muscle dysfunctions and acquired compensatory orofacial behaviors which may fall under the purview of myofunctional therapists[45]. The drowsiness and impaired alertness associated with untreated OSA has also

been shown to increase the risk of work-related injuries and motor vehicle accidents in association with the sleep fragmentation induced by abnormal breathing events[34]. Obesity is a major risk factor for OSA and is seems to be responsible for the growing prevalence in the last decade[21,41].

Other signs and symptoms suggesting sleep disordered breathing, such as scalloping of the tongue, tongue thrust at rest, chronic open mouth posture, or a low tongue position at rest, might still not be familiar to many health care professionals[15,44].

In children, prevalence has been reported in the range of 2 to 6%, with a peak in preschool-aged children, which has been related to the hypertrophy of the tonsils as a major risk, also common at this age group[25]. Childhood OSA can lead to cognitive impairment, behavioral problems, growth retardation, and cardiovascular and metabolic complications[25].

Similar, in adolescents, OSA is also associated with a wide range of cognitive and behavioral problems, from attention disorders to depression, sometimes with risk-taking behavior and suicidal tendencies, cardiovascular and metabolic complications. Although risk factors and physiopathology are not the same for children and adults, some approaches seem to be similar, and there is growing evidence that untreated OSA children are at higher risk to develop OSA in the adult age[18,25,34].

Different repercussions and treatment approaches enhance the understanding of OSA as a disease with several phenotypes and endotypes, with the need for individualized targeted approaches for personal precision medicine. These phenotypes are related mainly to the four mechanisms exposed as the pathways for physiopathology of OSA[14].

The main cause for OSA is still linked to the narrow, crowded airway, as part of the first mechanism, the *impaired upper airway anatomy*. The main clinical findings are associated with obesity, craniofacial dysmorphisms, hypertrophy of the tonsils, and/or nasal structures. Besides these, the shape and the size of the pharynx will also be influenced by its viscoelastic properties, reported as the collapsibility of the pharynx during inspiration[26].

As for non-anatomical phenotypes, the mechanism for OSA is related to neural control, muscle responsiveness, and muscle effectiveness.

In some patients, OSA development will be associated with a third mechanism related to the respiratory control (loop gain), considering that the most important respiratory driver is the CO2 level which might present fluctuations due to the narrowing of the airway during sleep and higher airway resistance.

The fourth phenotyping mechanism is linked to the respiratory arousal threshold. An increased respiratory drive, as induced by increased pharyngeal pressure swings, will recruit the dilator muscles of the upper airway. Thus, in people with low muscle responsiveness, activation of the pharyngeal muscles occurs only with severe gas changes[26].

TREATMENT OPTIONS

Treatment approaches are different for the pediatric and adult populations.

In pediatric OSA, adenotonsillectomy has been the treatment of choice. However, more detailed, and polysomnographic evaluation after surgery has shown a high rate of OSA persistence, ranging from 30 to almost 50%, depending on the characteristics of the studied population, like obesity, craniofacial disorders, or if syndromic children, as Down syndrome, were included[17]. Thus, a step-by-step treatment approach has been proposed, including clinical treatment with anti-inflammatory medication, rapid maxillary expansion, and weight loss, associated or not with myofunctional therapy intervention for muscle strengthening[25]. Besides this, these different outcomes have highlighted the recognition of different phenotypes, with the so-called "adult phenotype" corresponding to the more obese child with a lower chance of treatment success with adenotonsillectomy, and the "adenoid" one, associated with the hypertrophy of the tonsils, and the long-face characteristics with a good response to the surgical treatment. This predictive phenotyping has been proposed to be included in the screening evaluation "Sleep Clinical Record," improving the accuracy of the traditional questionnaires[42].

In adults, the treatment of choice is the support of positive pressure, the CPAP. In the case of poor compliance or intolerance to CPAP, the oral mandibular advancement device (MAD) is an alternative. Its efficacy, comparable to that of CPAP, is attributed to higher adherence to MAD than to CPAP. MAD is also indicated as a first-line treatment for moderate OSAS without associated severe cardiovascular comorbidities[1,37]. In some patients with positional OSA, the wearing of an anti-decubitus device can be prescribed to prevent sleepers from rolling on their back. Education on sleep positions

should be part of the overall treatment as there are some sleep positions that may reduce OSA more than others, depending on the condition of the patient[33,43]. An adapted diet, a physical activity program, the reduction of sleeping pills, alcoholic beverages, and tobacco consumption, are also indicated as general measures, improving sleep hygiene[8]. Surgical treatment of adult OSA involves numerous proposals, including soft tissue volume reduction by pharyngeal surgeries, or by optimization of the nasal patency, as nasal valve surgery, septoplasty, and turbinoplasty, with limited efficacy[46]. The highest success rate is obtained by maxillomandibular advancement surgery[24]. Hypoglossal nerve stimulation surgery is aimed at the contraction of the genioglossus muscle, the main dilator muscle of the pharynx, to suppress pharyngeal collapse during sleep[38].

OROFACIAL MYOFUNCTIONAL THERAPY

Within this short overview, there is growing evidence that orofacial structures and functions can play an important role in these mechanisms. Exercising and strengthening the orofacial muscles may have a positive impact on craniofacial growth, development and its functions, like sucking, swallowing, chewing, articulation, and influencing breathing patterns[2]. The importance of functional balance for more optimal facial growth, the harmonic appearance of the face, and achievement of nasal breathing, including the position and tonus of the tongue, has been emphasized by the orthodontic pioneer Alfred Paul Rogers already in the early 20th century, proposing active exercises to improve these conditions[36]. He first introduced the term "orofacial myofunctional therapy" (OMT) into the medical literature as a means to describe what he thought was the critical nature of the position of the tongue, at rest and in movement, on craniofacial morphology and the ensuing orthodontic and occlusal stability. Many of the orofacial myofunctional disorders (OMDs) originate with insufficient habitual nasal breathing or mouth breathing and may impact treatments by orthodontists, dentists, dental hygienists, speech-language pathologists, and other professionals working in the orofacial area.

Orofacial Myofunctional Therapy is a comprehensive approach that begins with creating awareness of the reciprocal impact of OSA on the orofacial musculature and oronasal functions, and the importance of therapy based on the principle of neuroplasticity, in order to promote long lasting behavioral changes to the patterns of orofacial functions[3,13,35,40]. A comprehensive approach to OMT includes isotonic and isometric exercising of the lips, tongue, soft palate, and pharyngeal walls, aiming to increase muscle tone, endurance, and coordinated movements of orofacial, pharyngeal and peripharyngeal muscles to optimize nasal breathing, chewing and swallowing[2,10,11,26].

In 2009, the first randomized clinical trial (RCT) of OMT specifically in adult OSA patients had been published by Guimaraes et al.[19]. Since then, numerous studies have performed different treatment protocols in adult and pediatric OSA patients, but many have relied on the same or similar exercises used in Guimaraes et al., spotlighting a need further research into specificity of the exercises and the phenotypes that would best benefit. Recently, including new methods to improve treatment adherence, a first RCT enrolling only severe OSA patient and OMT being monitored by an app has been published by O`Connor-Reina et al.[30]

Several systematic reviews confirm that OMT reduces snoring, the apnea-hypopnea-index and/or OSA severity, oxygen desaturation, and daytime sleepiness, but also improves the quality of life and the adherence to CPAP treatment[2,4,6,7,9,39]. These improvements might result from better muscle responsiveness, muscle gain, the coordinated recruitment of different compartments of tongue and other pharyngeal muscles, with reduction of inspiratory flow limitations and subsequent arousals[26]. There is a growing interest in identifying phenotyping tools and the site of obstruction in clinical settings, to select targeted OMT exercises, along with a comprehensive assessment of signs and symptoms of SDB easily detected even without sophisticated tools. As it was mentioned previously, some well-established signs and symptoms of OSA, easy to detect by clinicians, that could also be considered orofacial myofunctional disorders are tongue scalloping, a chronic open mouth posture with low tongue position at rest, a high score on the Mallampati or the Friedman scales, a tongue thrust at rest and during swallowing and/or speech, or visibly hypertrophic tonsils.

Not all trials for OMT have included a comprehensive or a validated assessment of myofunctional evaluation before and after executing their protocol. In the pediatric population, the combination of the orofacial myofunctional evaluation and a validated screening questionnaire improved the accuracy of the OSA diagnosis[12]. O'Connor-Reina et al. evaluated the Iowa Oral Performance Instrument (IOPI) to aid in the identification of the obstruction site in OSA patients. The authors observed a significant correlation of IOPI tongue pressures and VOTE classification (T size) during drug-induced sleep endoscopy[31].

Despite the availability of several validated protocols, such as the OMES-expanded, (which is validated for OSA patients[16]) or the MGBR[27] (which is not additionally validated for OSA patients) for evaluation, there is still a large variability in both research and clinical practice. There is also no consensus regarding therapy strategies.

Thus, the validation of OMT by meta-analysis has been challenging as data cannot be easily expanded to include different treatment protocols and patient characteristics due to lack of homogeneity and objective measurements on the effectiveness of the exercises themselves. Although the quality of evidence may be considered low due to limitations of the studies, all systematic reviews point out a positive effect of OMT, giving promise of potential for an impactful, emerging new field in sleep medicine[29]. Each year new peer-reviewed articles are added to the current knowledge of OMT's effectiveness, especially in treating patients with OSAS.

PROPOSAL TO STANDARDISE PROTOCOLS FOR ASSESSMENT AND MYOFUNCTIONAL REHABILITATION IN PATIENTS WITH OSA

Given the increased application of orofacial myofunctional sciences into therapy for a wide variety of disorders and conditions, it seems desirable to develop good practice guidelines. These are defined as "methodically developed proposals to assist the practitioner and the patient in seeking the most appropriate care in given clinical circumstances"[23]. They are intended to provide professionals, patients, and decision-makers in the health system with a rigorous summary of the state of the art, standardizing the assessment and treatment of orofacial myofunctional disorders in patients with OSA (children, adolescents, and adults).

Our approach aims to standardize the assessment and treatment of orofacial myofunctional disorders in patients with OSA (children, adolescents, and adults).

To develop recommendations for good practice, we chose to use a consensus method. We organized a meeting of a group of experts with the objective of developing a protocol of guidelines for assessment and treatment with RMOF. Of the four consensus methods classically described[22], we have chosen the Delphi method because it brings stakeholders together and systematically attempts to identify differences of opinion and facilitate consensus[20,28,32].

The Delphi method (fig. 1) is a group decision-making technique. It is based on feedback, after analyzing the results of questionnaires sent to a panel of experts. Their respective answers and references are summarized and forwarded to them for further comments. Consensus can be reached within a few rounds of this process. The Delphi technique reduces bias in data processing and prevents one person from unduly influencing the outcome. Critical success factors for its implementation are mainly effective facilitation, careful selection of experts and prior definition of target questions.

The results based on this process will help to develop an international expert-based consensus of recommendations for the protocol of orofacial myofunctional evaluation and treatment. The goal is to create a uniform base for the evaluation and the treatment of OSA in children and adults, and, thus, improve the quality of care and reduce variation in myofunctional therapy for OSA.

THE DELPHI PROCESS

The Delphi Consensus Statement on Orofacial Myofunctional Assessment and Treatment for Patients with OSA was initiated by Marc Richard Moeller, Executive Director of the Academy of Applied Myofunctional Sciences (AAMS). A variety of international professionals and researchers, with demonstrated expertise from relevant disciplines, have been included in the process and are providing ongoing feedback on the different steps and resources necessary to meet the objectives and the final goals.

The first two steps of the consensus recommendations Delphi consensus recommendations, the explicit definition of the subject Delphi method, and the selection of experts, have been completed.

The third stage, the development of the questionnaire, will be reviewed at the Academy of Applied Myofunctional Sciences/ World Sleep Special Interest Group Conference held in conjunction with the World Sleep Congress 2022 in Rome, Italy, March 2022.

The conclusions of the Delphi consensus recommendations will be published in the following year and widely disseminated.

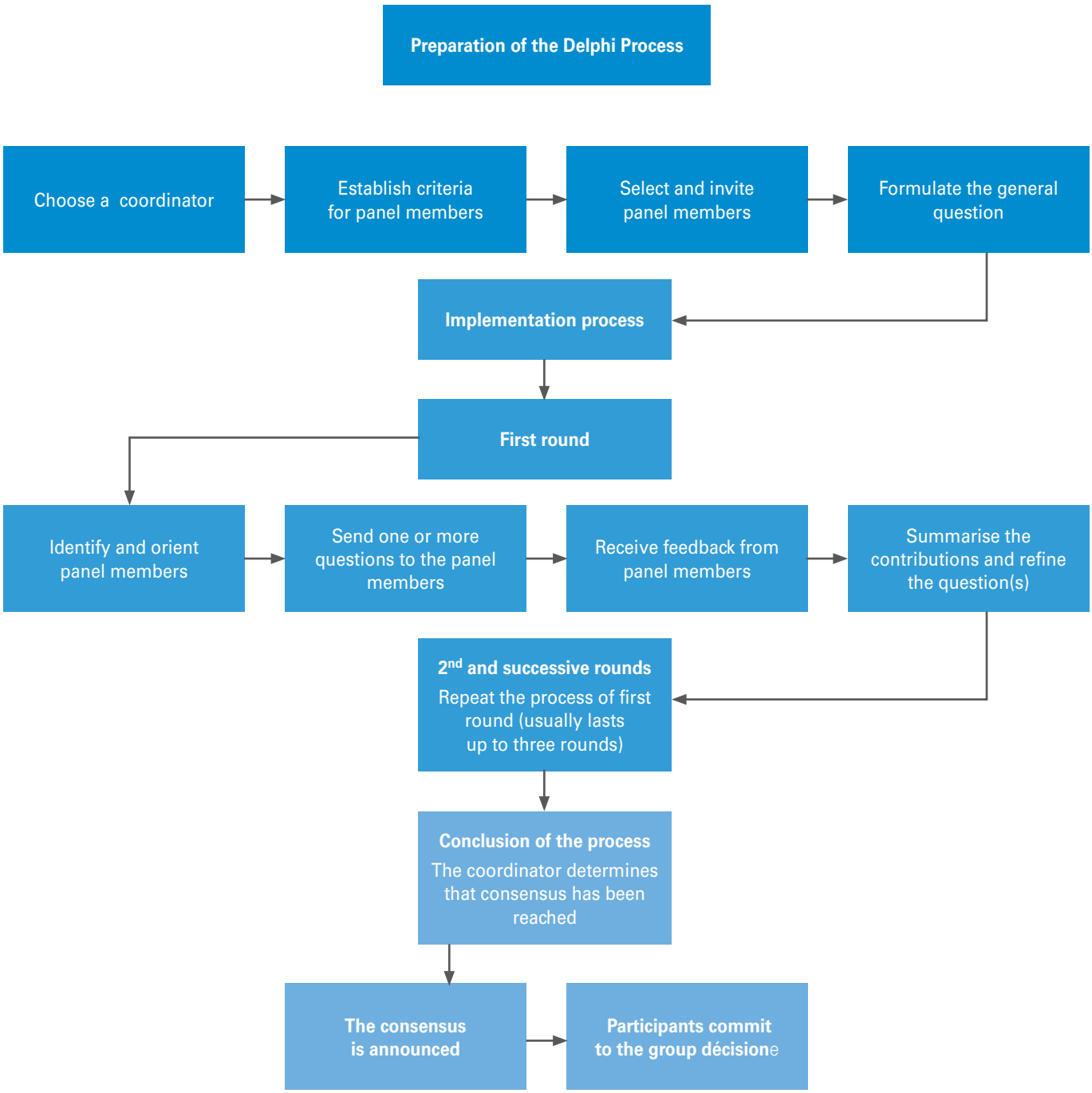

Figure 1: steps of the Delphi Process.

Conflicts of Interest:

The authors declare no conflicts of interest.

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All authors have read and agreed to the published version of the manuscript.

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