A Comparison of the Effects of Myofunctional Therapy Combined with Frenectomy on Satisfaction, Quality of Life, and Symptoms of Obstructive Sleep Apnea and Ankyloglossia in Adult Patients with Obstructive Sleep Apnea and Ankyloglossia: A Preliminary Study

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Abstract

Myofunctional therapy is a noninvasive therapeutic option for obstructive sleep apnea (OSA) patients, frequently provided in recent years because it is less expensive and has lower serious side effects compared to other methods. It can also be used to treat patients who are intolerant to continuous positive airway pressure (CPAP) or oral appliances. Ankyloglossia is a congenital condition that limits the movement of the tongue and may interfere with exercise regimens, thus reducing the efficiency of myofunctional therapy. The purpose of this study is to explore the effectiveness of myofunctional therapy combined with frenectomy in adult OSA patients with ankyloglossia.

In this prospective, cohort study, 15 adult patients aged 20 to 60 years and newly diagnosed with mild to moderate OSA and ankyloglossia were recruited and randomized into either a control group (myofunctional therapy alone) or an intervention group (myofunctional therapy combined with frenectomy). After 3 months of therapy, the outcomes were evaluated using the Epworth Sleepiness Scale (ESS), the patient-reported experience measures (PREMs), and the patient-reported outcome measures (PROMs) questionnaires.

Comparing ESS at baseline (T0) and 3 months after therapy (T3), there was a significant reduction of the ESS in both the control group (2.88±1.73) and intervention group (4.00±3.65). However, the decrease in ESS was not statistically different between the groups. Overall satisfaction in the control and intervention groups was 100% and 57.2%, and improvement in quality of life was 75.0% and 57.1%, respectively. The symptoms associated with OSA and ankyloglossia were improved in all categories for both groups.

It can be concluded that both myofunctional therapy alone and myofunctional therapy combined with frenectomy can improve ESS, quality of life, symptoms associated with OSA and ankyloglossia, and the ability to perform myofunctional therapy in adult patients with mild to moderate OSA and ankyloglossia although the reduction of ESS between the groups was not statistically different. However, according to the results, patients in the myofunctional therapy alone group had more satisfaction and more quality-of-life improvement.

Keywords: *Obstructive Sleep Apnea, Myofunctional Therapy, Tongue Exercise, Ankyloglossia, Frenectomy*

1. Introduction

Obstructive sleep apnea (OSA) is a syndrome characterized by frequent upper airway collapse that results in stoppage (apnea) or reduction of breathing (hypopnea) during sleep. Patients commonly have trouble sleeping, snoring, daytime sleepiness, and insufficient sleep (Marcus et al., 2012; Thorpy, 2012). These symptoms increase the risk of traffic accidents (Tregear, Reston, Schoelles, & Phillips, 2009) and occupational injuries (Allen et al., 2016). OSA also affects adult mortality by raising the risk of metabolic and cardiovascular diseases (Butler et al., 2019; Friedman & Logan, 2009).

The oropharyngeal region's collapsible portions, which comprise several muscles and soft tissue, are primarily responsible for the collapse of the upper airways. These muscle groups cooperate to perform daily activities including speaking, swallowing, and breathing. However, a reduction in muscle activity while sleeping may obstruct the upper airway and make it more collapsible. When apnea or hypopnea occurs, the sympathetic nervous system is activated, causing transitory blood gas disturbances including hypoxemia and

hypercapnia. Patients are awakened from sleep by arousal to reopen the upper airway. This incident causes fragmented sleep and frequent switching between awakening and sleeping, directly impacting OSA patients' daytime drowsiness and quality of life (Eckert & Malhotra, 2008; Patil, Schneider, Schwartz, & Smith, 2010).

Surgical and non-surgical therapies are available as therapeutic options for adult patients. Surgery aims to correct structural issues with procedures including Uvulopalatopharyngoplasty (UPPP), maxillomandibular advancement (MMA), and genial advancement. For the majority of patients, especially those with severe problems, continuous positive airway pressure (CPAP) has traditionally been the first-line treatment among non-surgical therapies. In mild to moderate cases, oral appliances, such as the tongue-stabilizing devices and the mandibular advancement devices (MAD), can be applied (Conley, 2011).

The tongue is frequently mentioned as one of the organs involved in the pathophysiology of OSA in the head and neck region. It was revealed that larger tongue volume and area, which are mostly caused by increased tongue fat and usually accompanied by decreased genioglossus muscle activity while patients sleep, had a major impact on upper airway collapse (Hirata et al., 2016; McSharry et al., 2014). Ankyloglossia is a congenital condition that limits tongue movement caused by a short lingual frenum. Patients with ankyloglossia often struggle to raise, protrude, and move the tongue laterally. Therefore, it might interfere with exercise regimens and reduce the efficiency of myofunctional therapy (Zaghi et al., 2019). Complete removal of restricted frenum (frenectomy) or incision and repositioning of frenum (frenotomy) can be performed to alleviate the problems caused by ankyloglossia, including tongue movement limitation, speech and feeding problems as well as maldevelopment of the dental arch. These surgical procedures can be performed using several instruments including a scalpel, electrosurgery, and laser (American Academy of Pediatric Dentistry, 2022; Devishree et al., 2012).

In recent years, myofunctional therapy has been provided as a therapeutic option for OSA in order to improve muscle weakness and dysfunction by increasing muscle coordination, tonicity, strength, and endurance as well as decreasing volume and fat in the muscles. This intervention includes isotonic and isometric exercises for the oropharyngeal area involving the tongue, pharynx, and facial muscles, which can reduce the severity of OSA and improve the quality of life in adult patients (de Felício, da Silva Dias, & Trawitzki, 2018). Myofunctional therapy is superior to other interventions for treating OSA because it is a noninvasive treatment, less expensive, and has no serious side effects. Furthermore, it is applicable to patients who are intolerant to CPAP or oral appliances (Rueda, Mugueta-Aguinaga, Vilaró, & Rueda-Etxebarria, 2019).

Effects of myofunctional therapy on OSA were discussed in numerous studies and reviews (Camacho et al., 2017; Diaferia et al., 2013; de Felício et al., 2018; Guimarães, Drager, Genta, Marcondes, & Lorenzi-Filhoy, 2009; Rueda et al., 2019). However, there are currently few investigations that focus on myofunctional therapy combined with surgical release of ankyloglossia (frenectomy) (Zaghi et al., 2019). Therefore, this study aims to explore the subjective outcomes including satisfaction and quality of life of myofunctional therapy combined with frenectomy in adult OSA patients with ankyloglossia.

2. Objectives

1) To compare the difference between ESS after myofunctional therapy with frenectomy and myofunctional therapy alone as the treatment for adult patients with obstructive sleep apnea and ankyloglossia.

2) To evaluate the patient's satisfaction after myofunctional therapy with frenectomy and myofunctional therapy alone as the treatment for adult patients with obstructive sleep apnea and ankyloglossia.

3) To evaluate the patient's quality of life after myofunctional therapy with frenectomy and myofunctional therapy alone as the treatment for adult patients with obstructive sleep apnea and ankyloglossia.

4) To evaluate the improvement of symptoms after myofunctional therapy with frenectomy and myofunctional therapy alone as the treatment for adult patients with obstructive sleep apnea and ankyloglossia.

5) To evaluate the improvement of the ability to perform myofunctional therapy after myofunctional therapy with frenectomy and myofunctional therapy alone as the treatment for adult patients with obstructive sleep apnea and ankyloglossia.

3. Materials and Methods

This prospective, cohort study was based on patients who presented at the Center for Sleep Disorders, King Chulalongkorn Memorial Hospital between January 1, 2022, and December 31, 2022.

3.1 Participant recruitment

Fifteen Thai adult patients, aged 20 to 60 years and newly diagnosed with mild to moderate OSA based on polysomnography interpretation by certified physicians and with ankyloglossia tongue range of motion ratio (TRMR) grade 2 to 4 by the dentist, were recruited in the study. TRMR is the ratio of maximal interincisal mouth opening (MIO) and mouth opening with tongue tip to maxillary incisive papilla at the roof of the mouth (MOTTIP), which can be categorized as grade 1 (> 80%), grade 2 (50-80%), grade 3 (<50%) and grade 4 (< 25%). All individuals must not have a body mass index (BMI) over 35, not undergo other OSA treatments, not present with uncontrolled systemic diseases, not present with mental retardation or psychiatric problems, not present with connective tissue or muscular disorders, not have a history of frenectomy or myofunctional therapy, not have a history of craniofacial trauma or oral and maxillofacial surgery (except extraction or surgical removal), and not have a large torus mandibularis which affects tongue movement.

Patients who met all inclusion criteria were given participant information sheets and informed by the researcher at the Center for Sleep Disorders about the study's details, including the study design, risks, and benefits. The patients interested in participating in the study were given an appointment at the Chulalongkorn University's Faculty of Dentistry for further discussion regarding the study. Before the study began, the patients were requested to sign the consent form. The study was approved by the Human Research Ethics Committee of the Faculty of Dentistry, Chulalongkorn University, Bangkok, Thailand (HREC-DCU 2021-027) and the Human Research Ethics Committee of the Faculty of Medicine, Chulalongkorn University, Bangkok, Thailand (COA No. 1428/2021)

3.2 Sample allocation

The patients were divided into the intervention group and the control group using block randomization generated by a digital block randomization generator (1:1 ratio). The patients with number 1 in the block were assigned to the control group and the patients with number 2 in the block were assigned to the intervention group.

Control group: Myofunctional therapy alone

In this group, the patients received myofunctional therapy and exercise instructions from a researcher. The patients were informed to exercise 3 times a day for 3 months.

Intervention group: Myofunctional therapy combined with frenectomy

From the same researcher, the patients in this group received myofunctional therapy and exercise instructions similar to the control group for 1 month. Then, the patients underwent frenectomy by an experienced maxillofacial surgeon. After the operation, the patients continued the exercise at home for 2 months, beginning the day after the surgery.

3.3 Intervention

Myofunctional therapy

The patients in both groups received myofunctional therapy instructions from the same researcher, consisting of 3 tongue exercises. The patients were informed to do all the 3 tongue exercises 3 times a day, in the morning, at noon, and in the evening. Patients also received the checklist notebook that included a tongue myofunctional therapy guidelines in one day and a personal checklist record.

1) Palate scrapes: Raise the tip of the tongue up to touch the most anterior point of the hard palate, then pull it back as much as possible and hold for 5 seconds; repeat 5 times.

2) Elephant swing: Open the mouth at resting position and protrude the tongue forward as much as possible, then move it to the left and right corner of the lip; repeat 5 times.

3) Tongue clicks: Elevate the tip of the tongue to touch the most anterior point of the hard palate like sucking with the palate, then clatter down with a 'click' sound; repeat 5 times.

All patients were provided with myofunctional therapy video instruction and asked to do the myofunctional therapy to ensure position and movement. Then, they were contacted by telephone after 2 months of treatment to follow up and improve patients' compliance.

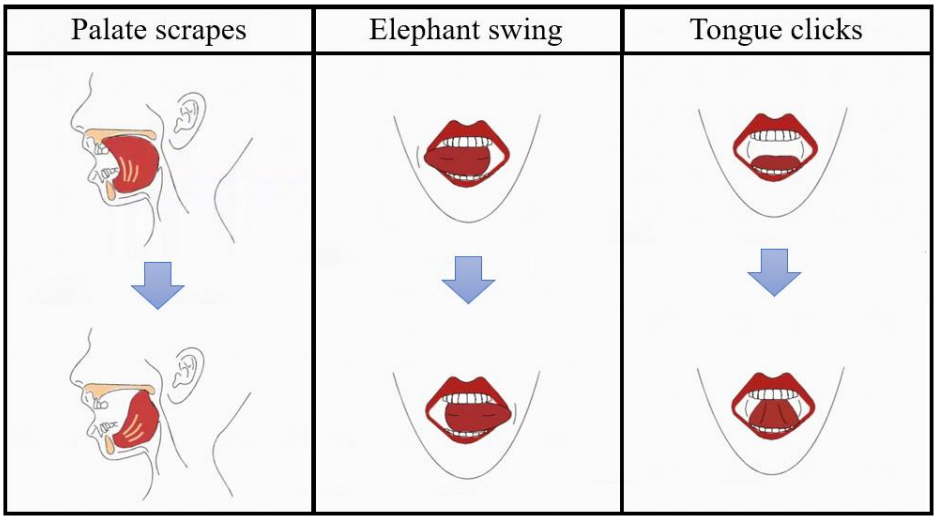

Figure 1 Myofunctional therapy instructions

Frenectomy

The patients in the intervention group underwent a conventional frenectomy technique by an experienced surgeon under local anesthesia. The surgical technique used 2% mepivacaine with 1:100,000 epinephrine, scalpel No. 15, and a hemostats approach. A curved hemostat was placed against the tissue facing the tongue's ventral surface, over the superior aspect of the frenum, and with its tips meeting the profound aspect near the base of the tongue. Then another curved hemostat was placed at the frenum tissue facing the floor of the mouth. The incision was made following the hemostats, blunt dissection of the frenum until the tongue reached the vermillion border of the lower lip. Afterward, the wound was carefully reapproximated and sutured with Vicryl suture 4-0. The postoperative period to follow up and stitch off was 7 days after frenectomy.

3.4 Measurement

The Epworth Sleepiness Scale (ESS)

Prior to the beginning of the treatment (T0), the ESS was measured by asking the patients to read and complete questionnaires in Thai. After 3 months of the treatment (T3), the ESS was re-measured and recorded to evaluate the treatment outcomes.

Patient-reported experience measures (PREMs) and patient-reported outcome measures (PROMs) questionnaires

The questionnaires were assessed with a 5-point Likert scale, consisting of questions about overall satisfaction, quality of life, symptoms of OSA and ankyloglossia, and the ability to perform myofunctional therapy. The questionnaires were surveyed and recorded after 3 months of treatment (T3) to evaluate the treatment outcomes.

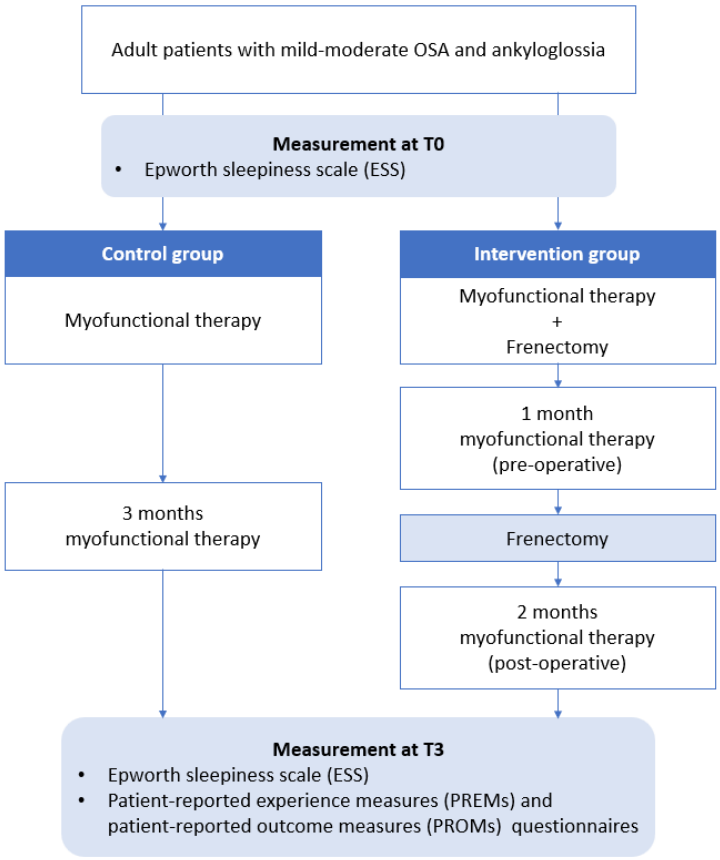

Figure 2 Study design

3.5 Statistical analysis

The Shapiro-Wilk test was used to assess the normality of data. The continuous data were summarized as mean (M) ± standard deviation (SD). The categorical data were summarized as frequency and percentages. The Mann-Whitney U Test was used to compare the ESS between the groups. The Wilcoxon Signed Rank Test was used to compare the ESS before and after the treatment. The p-values <0.05 were considered significant. All statistical analyses were performed using SPSS for Windows version 22.

4. Results and Discussion

4.1 Results

Table 1 Baseline demographic data

Variables	Control group (N=8)	Intervention group (N=7)	*p*-value
Age (years)	38.88±10.30	41.43±9.40	.69
Gender, male (%)	7 (87.50)	5 (71.43)	.61
Gender, female (%)	1 (12.50)	2 (28.57)	.61
BMI (kg/m^2)	26.29±3.98	25.45±3.49	.78
Neck circumference (cm)	15.06±1.05	14.36±1.63	.40
AHI (event/hour)	19.15±5.55	15.61±9.68	.40
ESS at T0 (point)	9.00±4.50	9.14±3.71	.87

**Significant at p-value < .05*

AHI=Apnea-hypopnea index

Fifteen patients (12 males, 3 females) were enrolled. The baseline demographic data are provided in Table 1. No statistical differences were found between groups in any characteristics, including age, gender, BMI, neck circumference, AHI, and ESS at the baseline.

Table 2 Comparison of ESS

Variables	Control group (N=8)			Intervention group (N=7)		
	T0	T3	*p*-value	T0	T3	*p*-value
ESS (point)	9.0±4.504	6.13±3.18	.02*	9.14±3.716	5.14±0.90	.03*
Reduction of ESS (point)	2.88±1.73			4.00±3.65		.78

**Significant at p-value < .05*

ESS

By comparing ESS at T0 and T3, there were significant reductions in the ESS in both the control group and the intervention group. However, the reduction of ESS between the groups was not statistically different.

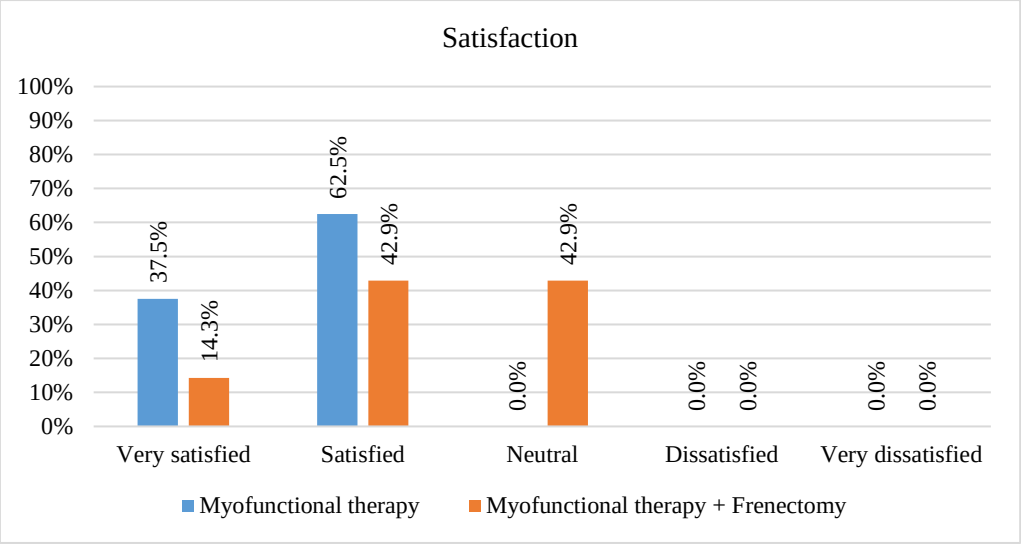

Figure 3 Percentage of patient's satisfaction in both groups

Satisfaction

There was 100% satisfaction with the treatment, including 37.5% of "very satisfied" and 62.5% of "satisfied", in the patients who received myofunctional therapy alone in the control group. In the intervention group, which combined myofunctional therapy with frenectomy, the patients reported 57.2% of satisfaction including 14.3% of "very satisfied" and 42.9% of "satisfied" Of these patients, 42.9% were reported neutral. No dissatisfaction was reported in any of the patients.

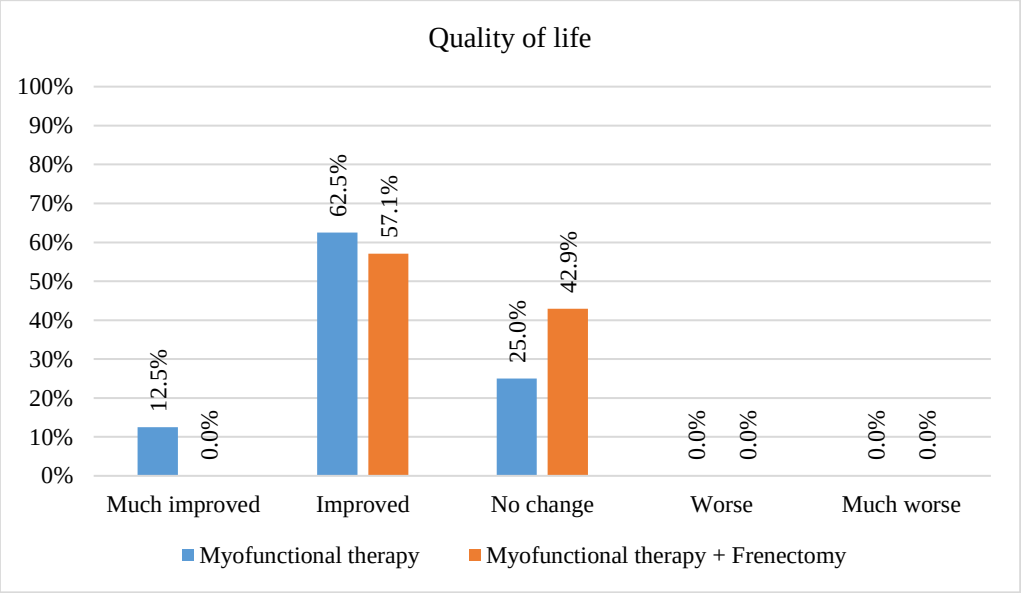

Figure 4 Percentage of patient's quality of life in both groups

Quality of life

The patients who received myofunctional therapy alone reported a 75.0% improvement in quality of life, including 15.5% of "much improved" and 62.5% of "improved", while the patients who received myofunctional therapy combined with frenectomy reported a 57.1% improvement in quality of life, all of which as "improved". No worsening in quality of life was reported in all patients.

Table 3 Symptoms involved with OSA and ankyloglossia in both groups

Myofunctional therapy						
Symptoms	Much improved	Improved	No change	Worse	Much worse	Improvement percentage
Nighttime sleeping	-	50.0% (4)	50.0% (4)	-	-	50.0%
Snoring	-	25.0% (2)	62.5% (5)	12.5% (1)	-	25.0%
Bruxism	-	37.5% (3)	62.5% (5)	-	-	37.5%
Nasal breathing	-	100.0% (8)	-	-	-	100.0%
Tongue movement	-	62.5% (5)	37.5% (3)	-	-	62.5%
Speaking	12.5% (1)	25.0% (2)	62.5% (5)	-	-	37.5%
Swallowing	12.5% (1)	50.0% (4)	37.5% (3)	-	-	62.5%
Head and neck tension	-	25.0% (2)	75.0% (6)	-	-	25.0%
Myofunctional therapy + Frenectomy						
Symptoms	Much improved	Improved	No change	Worse	Much worse	Improvement percentage
Nighttime sleeping	-	14.3% (1)	85.7% (6)	-	-	14.3%
Snoring	-	28.6% (2)	71.4% (5)	-	-	28.6%
Bruxism	14.3% (1)	14.3% (1)	71.4% (5)	-	-	28.6%
Nasal breathing	-	42.9% (3)	57.1% (4)	-	-	42.9%
Tongue movement	28.6% (2)	28.6% (2)	42.9% (3)	-	-	57.2%
Speaking	-	14.3% (1)	85.7% (6)	-	-	14.3%
Swallowing	14.3% (1)	14.3% (1)	71.4% (5)	-	-	28.6%
Head and neck tension	-	14.3% (1)	85.7% (6)	-	-	14.3%

Symptoms involved with OSA and ankyloglossia

For the patients who received myofunctional therapy alone, the improvement percentage was 100% in nasal breathing and 62.5% in tongue movement and swallowing. There was a patient that reported worsening snoring in this group. For the patients who received myofunctional therapy combined with frenectomy, the improvement percentage was 57.2% in tongue movement and 42.9% in nasal breathing. There was no report of the symptoms worsening in this group.

Ability to perform myofunctional therapy

There was 100% improvement in the ability to perform myofunctional therapy reported by the patients in both groups. The improvement in the intervention group included 42.9% of "much improved" and 57.1% of "improved", while the patients in the control group reported 12.5% of "much improved" and 87.5% of "improved".

4.2 Discussion

Myofunctional therapy is a treatment modality that involves isotonic and isometric exercises focusing on facial muscles, pharynx, and tongue. The purpose of this treatment is to alter muscle dysfunction and weakness by increasing coordination, tonicity, strength, and endurance of the muscles. In 2009, Guimaraes and colleagues introduced myofunctional therapy as a treatment for OSA (Guimarães et al., 2009). Since then, the effects of myofunctional therapy on OSA patients have been investigated and discussed in numerous studies. The systematic review and meta-analysis suggested that myofunctional therapy can decrease AHI and ESS, thus improving sleep quality and quality of life (Camacho et al., 2017; de Felício et al., 2018). Although CPAP is considered the first-line treatment for patients with OSA, especially in severe cases (Jordan, McSharry, & Atul, 2014), long-term compliance is still low (Afsharpaima, Shahverdi, Vahedi & Aqaee, 2020). However, myofunctional therapy is superior to other interventions for treating OSA because it is noninvasive, less expensive, and has no serious side effects. Furthermore, it can apply to patients who are intolerant to CPAP or oral appliances (Rueda et al., 2019).

The genioglossus muscle is the largest upper airway dilator muscle. The functions of the genioglossus muscle are tongue protruding and depressing. Besides, it plays an important role in dilating and strengthening the upper airway (Messina, 2017). It was suggested that increased tongue volume and area, which are mainly determined by more tongue fat as well as decreased genioglossus muscle activity, are responsible for OSA's pathogenesis and OSA's severity (Cori, O'donoghue, & Jordan, 2018). Moreover, the authors hypothesize that surgical release of the short lingual frenum, which restricts movement, can enhance the effects of myofunctional therapy. The degree of ankyloglossia can be measured using various methods, including the Kotlow free-tongue measurement, the Marchensan tongue range of motion deficit (TRMD), and the tongue range of motion ratio (TRMR). It was suggested to use TRMR for measuring the degree of ankyloglossia by the study focusing on validating grading scales for ankyloglossia, as it is an independent measurement method that is directly related to restrictions in the tongue function (Yoon et al., 2017).

The amount of time taken and complexity of the therapy were considered to decrease adherence to myofunctional therapy (Koka et al., 2021). To be more specific to the tongue and increase adherence to the therapy, the myofunctional therapy protocol in this study thus focuses on short time protocols that improve tongue tone, strength, and endurance. Pain after surgery may be an obstacle to practicing myofunctional therapy. To perform correctly after surgery, participants were therefore taught to exercise their tongue 1 month before surgery to familiarize the myofunctional program and improve their dexterity. After that, 2-month postoperative myofunctional therapy was instructed to stretch the wound to reduce the scar and enhance wound healing. Although pain may reduce the ability to do myofunctional therapy in a few days after surgery, the benefits of post-operative myofunctional therapy are still crucial.

Daytime sleepiness is considered a major symptom of OSA, which can lead to occupational injuries (Allen et al., 2016) and traffic accidents (Tregear et al., 2009) generally evaluated by ESS. The ESS was introduced by Johns since 1991 (Johns, 1991). The study suggested that this questionnaire can be used to follow up on the improvement of treatment in patients with OSA as it has excellent internal consistency and reliability. Furthermore, it can indicate the actual sleepiness in daily activities that cannot be shown in

laboratory environments in polysomnography (Banhiran, Assanasen, Nopmaneejumruslers, & Metheetrairut, 2011). However, the ESS is a subjective measurement, and thus it may involve individual psychology and bias. The results of this study reveal that ESS was reduced significantly in both groups of patients from T0 to T3 (9.0±4.50 to 6.13±3.18 in the control group and 9.14±3.71 to 5.14±0.90 in the intervention group, p=0.05), which is in good agreement with a previous study in 2022 suggesting that the tongue muscle training can significantly reduce the ESS in the intervention group (Poncin et al., 2022). The reduction of ESS between both groups (2.88±1.73 in the control group and 4.00±3.65 in the intervention group) had no statistical difference. However, this may be caused by the sample size limitation.

The percentage of satisfaction in this study was higher in the patients who received myofunctional therapy alone. There was 100% satisfaction with the treatment (37.5% of "very satisfied" and 62.5% of "satisfied"), while in the intervention group, patients who received myofunctional therapy combined with frenectomy had 57.2% satisfaction (14.3% of "very satisfied" and 42.9% of "satisfied"). The lower satisfaction in the latter group can be explained by the patient's pain and discomfort within a few days after undergoing frenectomy, as well as more follow-up visits for stitch off. Corresponding with the satisfaction result, the improvement percentage in quality of life was higher in patients who received myofunctional therapy alone, 75.0% in the control group compared to 57.2% in the intervention group. Neither group of patients reported dissatisfaction or worsening quality of life. By comparing the results from a study that explored comparable patients, our study reported slightly lower satisfaction and quality of life in the patients of the intervention group (Zaghi et al., 2019).

The improvement of nasal breathing was 100% in the control group, and 42.9% in the intervention group. Because myofunctional therapy may have the ability to restore the tongue position to rest against the palate, it can decrease mouth breathing and increase nasal breathing. Tongue movement reported by the patients was improved in both groups. However, the control group had more percentage of tongue movement improvement (62.5%) than the intervention group (57.2%). The improvement may be explained by increasing tongue strength and endurance which are aims of the myofunctional therapy. Both nasal breathing and tongue mobility improvement, which were found to be less in the intervention group, may have been attributed to surgical pain. As a result, the participants felt uncomfortable overall.

Focusing on the ability to perform myofunctional therapy, the patients in both groups reported 100% improvement in the ability to perform myofunctional therapy. The improvement in the intervention group included 42.9% of "much improved", while the patients in the control group reported 12.5%. The results corresponded with the hypothesis that surgical release of ankyloglossia can improve tongue mobility and the ability to do myofunctional therapy following the protocol in the intervention group.

The main limitation of the study is the small sample size. Due to the limited study time, the situation of COVID-19, and the exclusion of severe OSA patients, we could not recruit patients to meet the sample size target. Lack of objective measurement and a sham protocol are other limitations. For further studies, we suggest investigating more participants and follow-ups over a longer time period to explore long-term outcomes and compare the results. To reduce the bias from the questionnaire, we suggest handing out the questionnaire to one blind researcher and informing the participants that their answers can be anonymous.

5. Conclusion

Both myofunctional therapy alone and myofunctional therapy combined with frenectomy can improve ESS, quality of life, and symptoms associated with OSA and ankyloglossia in adult patients with mild to moderate OSA and ankyloglossia. However, the reduction of ESS between the groups was not statistically different. According to the results, patients in the myofunctional therapy alone group had more satisfaction and more quality of life improvement. Further studies with a larger group of participants and a longer follow-up period are needed to confirm the results.

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