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Efficacy of oral myofunctional therapy in middle-aged to elderly patients with obstructive sleep apnoea treated with continuous positive airway pressure

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Abstract

Background: Oral myofunctional therapy (MFT) is an effective treatment for mild-to-moderate obstructive sleep apnoea (OSA) in middle-aged patients. However, few reports have described its use in elderly patients with moderate and severe OSA. Moreover, no studies have examined the relationship between changes in tongue pressure with MFT and the severity of OSA.

Objective: We conducted an interventional study using MFT to evaluate the effect of MFT on middle-to-senior-aged patients with moderate or severe OSA and compared changes in apnoea-hypopnea index (AHI) and tongue pressure.

Methods: Thirty-two OSA patients (≥45 years) treated with continuous positive airway pressure (CPAP) were included. MFT was performed in parallel with CPAP. Three days after CPAP discontinuation, polysomnographies were performed and tongue pressures were measured before and after MFT.

Results: Patients were 69.3 ± 1.5 years old. After 6 months of MFT, AHI decreased significantly from 34.7 to 29.0/h ($P = .03$), while tongue pressure significantly increased from 35.9 to 45.6 kPa ($P < .01$). Seven patients (22%), including 6 of the 12 patients with moderate OSA (50%), experienced successful CPAP discontinuation.

Conclusions: MFT can be a useful intervention even among middle-aged to elderly patients with OSA. Increased tongue pressure may have contributed to the AHI improvement.

Clinical trials: Trial registration at www.umin.ac.jp UMIN000027547.

KEYWORDS
continuous positive airway pressure, middle-to-senior, myofunctional therapy, polysomnography, sleep apnoea, tongue pressure

1 | INTRODUCTION

Obstructive sleep apnoea (OSA) is a common breathing disorder characterised by repetitive narrowing and closure of the upper airways during sleep. OSA is associated with major adverse health outcomes, including increased cardiovascular, metabolic and cognitive issues. The prevalence of OSA increases with age, and 20% of people aged ≥ 70 years have OSA.[1] As a first-line treatment,

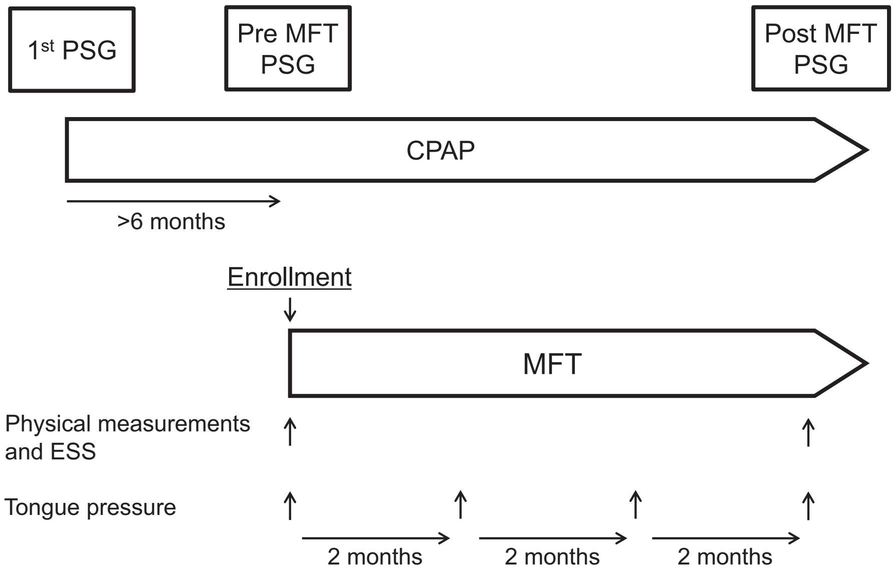

FIGURE 1 Time course of the study protocol. Patients were enrolled more than 6 mo after the initiation of continuous positive pressure (CPAP). Each polysomnography (PSG) was performed 3 d after discontinuation of CPAP, and treatment was resumed immediately after completion of the tests. Tongue pressure was measured every 2 mo to provide for feedback to the patients, but only results before and after oral myofunctional therapy (MFT) were used in the analysis, as with physical measurements and the Epworth Sleepiness Scale (ESS)

continuous positive airway pressure (CPAP) is highly effective in reducing sleep-disordered breathing events; however, approximately half of patients with OSA who attempt CPAP therapy have been reported to be either completely intolerant or only partially adherent.[2] Oral appliances (OA), which have a higher adherence than CPAP,[3] has also been widely used. Adjustable OA was effective among patients with moderate and severe OSA, but the evidence regarding the effects of OA on the long-term prognosis and prevention of cardiovascular events is still uncertain.[4] Furthermore, both CPAP and OA are symptomatic treatments, thus compensating for underlying issues; OSA symptoms will reappear when the patient stops using the devices. Therefore, new OSA treatments that address the causes of the disorder are clearly required.

When considering the pathophysiology of the disorder, an anatomically compromised or collapsible upper airway (high passive critical closing pressure of the upper airway) is considered the primary cause of OSA.[5] Alternatively, it has also been reported that one or more non-anatomical pathophysiological traits are present in 69% of patients with OSA.[6] Oral myofunctional therapy (MFT) is a non-anatomical approach for the treatment of OSA, aimed at functionally obtaining the appropriate positioning of the tongue: the tip of the tongue touches the hard palate behind the front teeth, allowing appropriate swallowing and breathing through the nose while the mouth is closed. A systematic review of 9 studies including a total of 120 middle-aged patients with mild-to-moderate OSA[7] demonstrated positive outcomes of MFT in several sleep parameters, including oxygen nadir, snoring, daytime sleepiness and improvement in apnoea-hypopnea index (AHI), which dropped by approximately 50%. However, few studies have reported the efficacy of MFT on middle- to– senior-aged patients with moderate and severe OSA.[7,8] In addition, it is not yet clear how MFT contributes to improving OSA. Tongue pressure is lower in young to middle-aged patients with severe OSA compared with healthy controls.[9] MFT may contribute to improvement in sleep-disordered breathing by increasing tongue pressure; however, the evidence has been limited to studies including children.[10]

In contrast to CPAP, MFT can be a fundamental treatment. Furthermore, investigating the effect of MFT is also important for patients with CPAP adherence issues. Thus, we performed a prospective study to investigate the effect of MFT among middle-to-senior-aged OSA patients with CPAP utilising polysomnography (PSG) and tongue pressure measurements, based on the hypothesis that MFT for relatively elderly patients can improve the severity of OSA.

2 | METHODS

2.1 | Patients

For this trial, we recruited ≥ 45-year-old patients with moderate-to-severe OSA through the Division of Comprehensive Sleep Medicine at the Tokyo Women's Medical University Hospital. These patients had been treated with CPAP for > 6 months. Because the effects of CPAP persist for a few days after discontinuation,[11] we performed full polysomnography (PSG) recordings 3 days after CPAP discontinuation. The PSG results obtained at OSA diagnosis were retrospectively confirmed. AHI did not change significantly with CPAP, and body weight did not vary significantly at inclusion (Figure S1).

We excluded patients with one or more of the following conditions: body mass index (BMI) ≥ 30 kg/m^2, craniofacial malformation, neuromuscular disease, previous stroke, heart failure or severe obstructive nasal disease. The Tokyo Women's Medical University Hospital Ethics Committee approved the study (approval number 170 204), and all patients provided written informed consent. Given the limited number of patients treated with CPAP in our hospital, we did not perform power analysis in the process of study design. All patients who consented to this intervention study were enrolled from April 2017 to October 2018.

Among the 41 patients enrolled in this study, 1 was excluded according to the exclusion criteria, 4 dropped out from the MFT programme during the 6-month training period, and 4 were excluded due to low adherence, as defined in Methods section 2.2. A total of 32 patients (22 males, aged 47-81 years) were included in the final analysis, with a majority of 23 (72%) being over 65 years of age. One patient was in their forties, 3 in their fifties (9%), 13 in their sixties (40%), 11 in their seventies (34%) and 4 in their eighties (13%). The mean age was 69.3 ± 1.5 years, and the mean body mass index was 23.9 ± 2.4 kg/m^2. Twenty patients (63%) had severe OSA.

2.2 | Study design

The experimental design is displayed in a flowchart (Figure 1). All patients were instructed by 2 dentists (T.O. and Y.A) to perform the MFT tasks described below and record their compliance in a diary. Tongue pressure was also measured 3 times using a disposable tongue pressure measurement device, with maximal tongue pressure applied.[12] The patients performed MFT 3 times a day at home for 6 months in parallel with CPAP treatment during sleep.[7,13,14] The patients visited the dentist every other month, and their MFT methods and compliance were confirmed. The results of the tongue pressure measurements were also reported back to the patients during each visit to reinforce their practice of MFT. In addition, PSG was performed before and after 6 months of MFT. Simultaneously, data on tongue pressure and subjective sleepiness (scored using the Epworth Sleepiness Scale [ESS]) before and after 6 months of MFT were collected for analysis. Patients with low compliance, i.e., those who performed MFT for less than 2/3rd of the treatment period, were excluded from the analysis. The primary outcome was AHI, and secondary outcomes included ESS, average apnoea event duration, AHI in the supine and lateral positions, AHI at the REM stage (REM AHI), minimal oxygen saturation (SpO2), percentage of time spent SpO2 < 90% with visually scored total sleep time, snore proportion and tongue pressure.

2.3 | PSG examination

A full-laboratory PSG examination was conducted overnight for each subject using Neurofax EEG-9200 (Nihon Kohden; Tokyo, Japan). Sleep status was determined based on the data from electroencephalography, electrooculography and submental electromyography, which were examined using the software included with the apparatus (Polysmith QP-260A, Neurotronics; Gainesville, FL, USA). Percutaneous oxyhemoglobin saturation (SpO2) was measured using a pulse oximeter (JL-951T3, Nihon Kohden; Tokyo, Japan). The presence of apnoea or hypopnea was determined by examining oral and nasal airflow in terms of thermocouples, nasal air pressure and thoracic and abdominal motions recorded using piezoelectric belt sensors. Simultaneously, a bipolar electrocardiogram (ECG) was recorded at a sampling frequency of 200 Hz to obtain the R-R interval (RRI) data during sleep. High and low pass filter settings of 0.1 and 100 Hz, respectively, were applied for ECG waveforms. The sleep stages and disturbed respiratory events were scored by a trained sleep technician based on the recommendations of the American Academy of Sleep Medicine. AHI, arousal index (total number of respiratory and non-respiratory arousals divided by the duration of sleep in hours) and minimal SpO2 were recorded. Snores were automatically scored by detecting bursts longer than 0.6 seconds on the snore channel, and the percentage of visually scored total sleep time spent in snore (snore %) was also determined. Moderate and severe OSA were defined by AHI values between 15.0/h and 29.9/h and ≥30.0/h, respectively.

2.4 | MFT tasks

We used our original MFT tasks based on OSA pathophysiology, as described in previous reports.[7,13,14]

2.4.1 | Tongue rotation exercise

The patient slowly rotates the tongue in the oral vestibule, clockwise and counterclockwise, 10 times each, 3 times a day, taking care not to open the lips during the exercise and using a hand mirror to check the execution.

2.4.2 | Cheek exercise

The patient repeats inflating and sucking in both cheeks 3 times each, 3 times a day.

2.4.3 | Pronunciation exercise

The patient pronounces 'Pa· Pa· Pa· Pa/Ta· Ta· Ta· Ta/Ka· Ka· Ka· Ka/La· La· La· La' 3 times slowly, 3 times a day.

2.4.4 | Nasal breathing rehabilitation

The patient develops the habit of breathing through the nose with the appropriate tongue position and specifically with the tip of the tongue touching the hard palate behind the front teeth, separating the teeth of the upper and lower jaws, while the lips are closed. The patient places several signs (eg a sign containing a star shape or a sign saying 'PAY ATTENTION TO YOUR TONGUE!') in his or her home or office as reminders to maintain a habit of breathing through the nose while maintaining the appropriate tongue position.

2.5 | Statistical analysis

Friedman tests were used to compare AHI and body weight; for the other variables, paired t tests were used to compare normally distributed variables and Wilcoxon tests or Mann-Whitney tests for skewed distributions pre- and post-MFT. Other than age, BMI, body weight, neck circumference and the ratio of time spent in the lateral position to total sleep time, the data for the remaining variabilities did not follow the assumption of normal distribution. All calculations were performed using SPSS 17 (SPSS Inc, Chicago, IL, USA). Statistical significance was set at $P < .05$.

3 | RESULTS

After 6 months of MFT, the AHI decreased significantly from 34.7 to 29.0 ($P = .03$, Figure 2), while the tongue pressure increased significantly from 35.9 to 45.6 ($P < .01$, Figure 3). There was no significant change in mean body weight from 64.3 ± 9.0 kg pre-MFT to 64.6 ± 9.2 kg post-MFT.

AHI in the lateral position decreased significantly (Table 1), while the ratio of time spent in the lateral position to total sleep time before and after MFT was 0.43 ± 0.2 and 0.46 ± 0.3, respectively, a non-significant difference.

Furthermore, average apnoea event duration and the ESS score decreased significantly (Table 1). In terms of OSA severity, 10 out of 22 patients with severe OSA (45%) improved to moderate status (n = 9) or mild (n = 1) OSA, and 7 out of 13 patients with moderate OSA (54%) improved to mild OSA status. Overall, reduction of OSA severity as assessed by AHI was observed in 26 out of 35 patients (74%). Particularly, OSA severity reduction was observed in 17 out of 25 (68%) patients aged ≥ 65 years.

Finally, 7 patients out of the total (22%), including 6 out of the 12 patients with moderate OSA (50%), experienced successful CPAP discontinuation due to their AHIs decreasing to values <20, at which point the Japanese social medical insurance scheme no longer covers the CPAP rental fee and patients must receive OA treatment instead. The median AHI value for these patients decreased from 20.9 (interquartile range, 16.8-25.0) before MFT to 11.9 (interquartile range, 9.9-13.9) afterwards ($P < .01$), and the median tongue pressure increased from 31.4 kPa (interquartile range, 28.9-33.9 kPa) to 47.5 kPa (interquartile range, 37.5-57.5 kPa) ($P < .01$).

Relative to the patients who stayed on CPAP after MFT, those who successfully discontinued CPAP had lower baseline BMI values (mean values, 22.2 ± 2.3 kg/m^2 vs 24.3 ± 2.5 kg/m^2; $P = .03$) and lesser baseline AHI severities (median values, 20.9/h [interquartile range, 16.8-25.0/h] vs 38.8/h [interquartile range, 27.8-49.8/h]; $P < .01$). As for sex distributions, 5 of the 7 patients who discontinued CPAP were female, but only 5 of the 25 patients who did not were female. The patients who discontinued CPAP and the patients who stayed on CPAP did not significantly differ in terms of age (mean values, 66.7 ± 9.0 years vs 69.5 ± 8.3 years).

4 | DISCUSSION

This is the first investigation of the efficacy of MFT on middle-to-senior-aged OSA patients with CPAP evaluated by full PSG and tongue pressure measurements. For the patients with OSA whose AHI did not change during the treatment with CPAP, 6 months of MFT significantly reduced AHI, resulting in 22% of the total, including 50% of moderate OSA patients achieving withdrawal

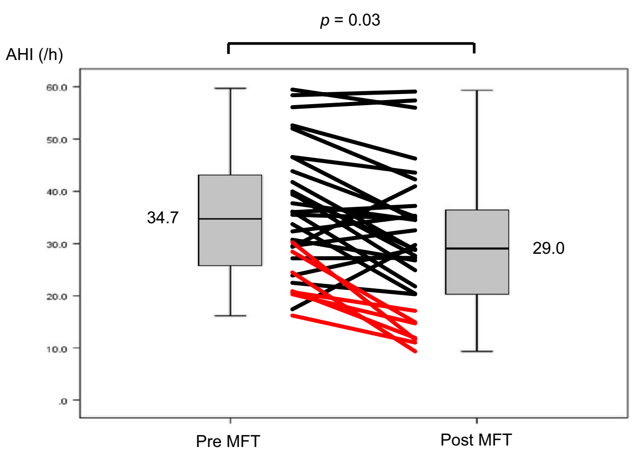

FIGURE 2 AHI changes following oral MFT. The boxplots show AHI distributions before and after 6 mo of MFT. The lines between the boxplots show AHI changes for individual patients, and the red lines represent patients who discontinued CPAP due to post-MFT AHI values < 20. Seven patients who achieved withdrawal from CPAP are identified by the red line (22%). AHI, apnoea-hypopnea index; CPAP, continuous positive airway pressure; MFT, myofunctional therapy

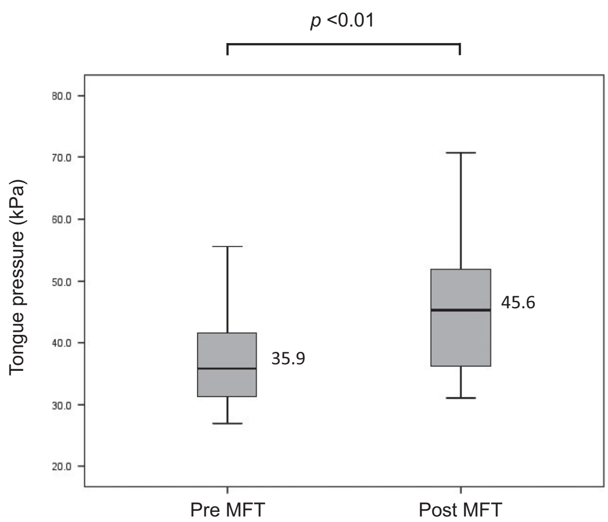

FIGURE 3 Tongue pressure changes following oral MFT. The boxplots show tongue pressures before and after 6 mo of MFT. The median tongue pressure increased from 35.9 (interquartile range, 30.1-41.7) before MFT to 45.6 (interquartile range, 37.8-53.8) afterwards ($P < .01$). MFT, myofunctional therapy

TABLE 1 Anthropometric and symptom data before and after 6-months MFT of whole patients (n = 32)

Variables	Pre-MFT	Post-MFT	*P* value
Body weight (kg)	64.3 ± 9.0	64.6 ± 9.2	.22
Neck circumference (cm)	37.9 ± 3.9	37.3 ± 3.8	.10
Abdominal circumference (cm)	89.5 (84.3-94.7)	89.3 (82.8-95.5)	.29
Apnoea index (/h)	14.3 (6.0-22.5)	8.1 (1.7-14.4)	**<.01**
AHI (/h)	34.7 (25.2-43.5)	29.0 (20.3-26.8)	**.03**
AHI in the lateral position (/h)	21.1 (8.6-33.6)	13.6 (7.8-19.3)	**.04**
AHI in the supine position (/h)	47.5 (38.5-57.0	46.7 (34.1-59.3)	.26
AHI at stage REM (REM AHI) (/h)	35.3 (26.9-44.2)	34.7 (20.4-49.0)	.60
Average apnoea event duration (s)	30.6 (24.6-46.6)	25.1 (19.8-30.4)	**.04**
Average hypopnea event duration (s)	23.8 (10.3-37.3)	24.5 (20.8-28.2)	.59
Arousal index (/h)	37.7 (27.0-48.4)	32.4 (24.3-40.5)	.53
Minimal SpO_2 (%)	80.0 (73.5-86.5)	81.0 (77.0-85.0)	.10
SpO_2 < 90% (%)	2.7 (0.0-7.5)	2.1 (0.0-6.0)	.15
Snore proportion (%)	1.9 (1.3-2.2)	2.0 (1.3-2.7)	.08
Epworth Sleepiness Scale (point)	6.0 (3.5-8.5)	5.0 (2.0-8.0)	**<.01**
Tongue pressure (kpa)	35.9 (30.1-41.7)	45.6 (37.8-53.4)	**<.01**

Note: Values in the table are expressed as means ± standard deviations or, in the cases of variables with skewed distributions, as medians (with interquartile ranges). Significant *P* values are indicated with bold text.
Abbreviations: AHI, apnoea-hypopnea index; MFT, myofunctional therapy; REM, rapid eye movement; SpO_2, oxygen saturation.

from CPAP without significant changes in body weight. A significant improvement in tongue pressure was observed at the same time, which may have contributed to the improvement in OSA. It should be noted that although tongue pressure decreases with age,[15] it has also been demonstrated that tongue pressure can improve after MFT even among middle-to-senior-aged patients. Increased tongue pressure, together with the styloglossus and genioglossus muscles may have contributed to the appropriate positioning of the tongue, that is the tip of the tongue touching the hard palate behind the front teeth.[14]

Average apnoea event duration was significantly reduced after MFT, possibly due to increased tongue muscle strength and the acquisition of the appropriate tongue positioning. AHI in the lateral position was also significantly reduced after MFT, while there was no significant change in the proportion of time spent in the lateral position during sleep before and after MFT. To date, there has been no study examining the effects of MFT on body position-dependent sleep-disordered breathing. The findings of this study suggest that MFT may affect breathing more in the lateral position than in the supine position, possibly due to the change in pharynx space.

Among the exercises in our study, tongue posture (maintaining appropriate tongue positioning) exercise was regarded as an important target, as it is fully justified by the findings related to the pathogenesis of OSA. The tongue rotation exercise and the pronunciation exercise, which were expected to increase the strength in the tongue, were also applied for the purpose of labial seal and of increasing the lip tone. Combined with the finding that the labial closure training alone did not significantly improve the symptoms of sleep-disordered breathing,[16] the result of the present study emphasises the importance of tongue exercise on OSA. Although upper airway collapsibility among OSA patients is associated with increased tongue volume,[17,18] whether MFT reduced the increased tongue volume was not examined in the present study. Future research on MFT should include assessment of not only tongue pressure, but also tongue volume.

In addition to the tongue exercise, nasal breathing rehabilitation is also important given that many patients with OSA have the habit of breathing through the open mouth from childhood. Such mouth breathing may be related to 'disuse' of the nose when breathing.[19] Intermittent hypoxia due to mouth breathing and/or sleep-disordered breathing has been reported to suppress the growth and development of the midfacial area, including the nasal cavity.[20,21] Thus, mouth breathing and sleep breathing disorders are locked in a vicious cycle. MFT can break the cycle from mouth breathing to sleep breathing disorders in adults, allowing for the achievement of nasal breathing with appropriate tongue position, as well as breaking this cycle for children with OSA.[22]

Cheek exercises were also included, which may improve symptoms by remodelling the oropharyngeal airway, increasing volume and flaccidity of the cheeks.[23]

Furthermore, although not included in this study's training regimen, the importance of diaphragmatic rehabilitation has been suggested,[14,24] based on the theory that tongue and diaphragm motor activity is controlled by the hypoglossal motor nucleus.[25] Since MFT involves a variety of training methods, among which intervention of MFT is most effective, and what mechanisms contribute to the improvement of sleep-disordered breathing should be clarified in future studies.

As for the intervention period, in previous studies,[8,14,16,22,24,26-28] the therapeutic programme lasted 2 or 3 months and included weekly visits with the speech therapist and/or home tasks. A critical difference in this study is the longer intervention duration than that in previous studies[8,14,16,22,24,26,27]; 6 months of intervention appears necessary to change behaviour through lifestyle intervention.[29] Compared to other studies,[8,14,24,26-28] the patients here required less frequent visits and no specific tool was required.[14,24] The patients were able to perform the exercises not only at home but also in their workplaces, with minimal time investment and lifestyle disruption. Meanwhile, it is still unclear how long the effects of the 6-month MFT intervention will last. Thus, to achieve more effective MFT, future investigations should consider both method and duration of the intervention, together with the duration of the effects.

Our study has some limitations. First, the participants were previously treated with CPAP and may have been accustomed to nasal breathing so that MFT exercises may have affected them more than patients not previously treated with CPAP. Second, many participants in this study could have been highly motivated to achieve complete remission of OSA to allow them to cease using CPAP; hence, their motivation to perform the MFT tasks could be higher than that of patients satisfied with CPAP treatment. Third, obese patients were excluded from the study. The apnoeic tongue in obese patients has a large volume of fat; therefore, it is uncertain whether MFT would be effective in obese older patients with severe OSA without being accompanied by weight reduction.

5 | CONCLUSIONS

Our data suggest that 6 months of MFT may have a beneficial effect for middle-aged to elderly OSA patients treated with CPAP. The increased tongue pressure observed after MFT may have contributed to the improvement of the severity of OSA. Therefore, the effect of MFT among middle-aged and elderly patients should be extensively examined in future prospective studies.

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CONFLICT OF INTEREST

Mayumi Suzuki reports Personal fees from TEIJIN, personal fees from MSD, personal fees from Medical Review KK outside the submitted work. Kentaro Matsui reports Personal fees from Eisai, personal fees from Meiji Seika Pharma, personal fees from Mochida, personal fees from MSD, personal fees from Otsuka, personal fees from Yoshitomiyakuhin, grants from Ministry of Education, Culture, Sports, Science and Technology, outside the submitted work. Haruki Sekiguchi reports Grants from National Hospital Organization, outside the submitted work. Nobuhisa Hagiwara reports Grants and Personal fees from Bayer Yakuhin, Ltd, grants and personal fees from Nippon Boehringer Ingelheim Co, Ltd, personal fees from Bristol-Myers Sqibb K.K, grants from Daiichi Sankyo Company, grants from Pfizer Japan Inc, outside the submitted work. The other authors declare no conflict of interest.

AUTHOR CONTRIBUTIONS

Mayumi Suzuki contributed to the conception and design of the study, acquisition, analysis and interpretation of data, original draft preparation and reviewing and editing the manuscript. Toshihiro Okamoto contributed to the conception and design of the study, acquisition of data and reviewing and editing the manuscript. Yuichi Akagi contributed to the acquisition of data. Kentaro Matsui contributed to the acquisition of data and reviewing and editing the manuscript. Haruki Sekiguchi contributed to the acquisition, analysis and interpretation of data and reviewing and editing the manuscript. Natsumi Satoya contributed to the acquisition of data. Yuji Inoue contributed to the conception and design of study, acquisition of data and reviewing and editing the manuscript. Akihisa Tatsuta contributed to the acquisition of data. Nobuhisa Hagiwara contributed to reviewing the manuscript and supervising the study.

PEER REVIEW

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SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section.

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