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〔Original Article〕

A pilot study of a two-arm, non-randomized controlled trial of guided self-help cognitive behavioral therapy for stuttering

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Abstract

To examine the effectiveness of guided self-help cognitive behavioral therapy（CBT）among adults who stutter when applied concomitantly to speech therapy, a pilot study was conducted on patients who were diagnosed as stuttering by an otolaryngologist. Patients were asked to choose between CBT and control groups. The CBT group received seven guided self-help CBT sessions once every one to two weeks, and four speech therapy sessions. Patients in the control group only received the latter once every three to four weeks. To measure subjective severity of stuttering, we used a visual analog scale. To measure stuttering problems, we used the Overall Assessment of the Speaker's Experience of Stuttering（OASES）and a fluency of speech measure; to measure social anxiety, we used the Liebowitz Social Anxiety Scale（LSAS）, the Social Phobia Inventory（SPIN）, and the Short Fear of Negative Evaluation scale（SFNE）; to measure depression, we used the Patient Health Questionnaire-9（PHQ-9）; to measure anxiety, we used the Generalized Anxiety Disorder-7（GAD-7）; to measure quality of life, we used the EQ-5D-5L. In total, 12 participants had their data analyzed. The mean reduction in the visual analog scale regarding subjective stuttering severity was non-significantly larger in the CBT group than in the control group, with a large effect size. The scores for the OASES, LSAS, SFNE, and GAD-7 of the CBT group showed larger effect sizes than those of the control group. Our findings suggest that guided self-help CBT for stuttering may improve subjective distress for stuttering and social anxiety.

***Key words*: cognitive behavioral therapy, stutter, speech therapy, social anxiety disorder, overall assessment of the speaker's experience of stuttering**

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Ⅰ. Introduction

According to the DSM-5[1], stuttering refers to a childhood-onset fluency disorder. Symptoms of stuttering include repetitions, prolongations of sounds, audible or silent blocking. Incidence rate approximately 8～11% of infants worldwide[2,3].

Howell and Davis[4]have created a model to predict whether eight-year old children will persist with their stuttering or recover by the time they become teenagers. Generally speaking, children who stutter are often teased and bullied by others[5], and these negative experiences can lead to mental health problems, including depression and social anxiety disorder (SAD)[6]. Moreover, such mental issues may be externalized by the difficulty of talking in front of others due to feeling extremely ashamed[7]. One study analyzed data from 28 community surveys from the World Mental Health Survey Initiative and included 142,405 respondents, aged 18 or older. This study found prevalence estimates of SAD over 30-day, 12-month, and the lifetime are 1.3, 2.4, and 4.0% across all countries[8]. Blumgart, Tran, and Craig[9]found that approximately 40% of the adults in their sample who stutter have SAD.

Among the possible treatments for SAD, which include selective serotonin reuptake inhibitors (SSRIs)[10], individual cognitive behavioral therapy (CBT) using the Clark and Wells model for SAD has been found to be the most effective[11]. A previous randomized controlled trial examined the effectiveness of CBT for SAD among participants who remain symptomatic despite receiving treatment with SSRIs, and showed that the addition of CBT helped reduce the severity of participants' SAD and depressive symptoms and helped improve their functioning and quality of life when compared to the control group[12,13].

To help understand the maintenance of stuttering behavior, the cognitive, affective, linguistic, motor, and social (CALMS) multi-dimensional model focuses on these five mentioned components of the lives of adults who stutter[14]. Nonetheless, the main treatment for children and adults who stutter remains speech therapy [15], which indeed improve the linguistic and motor factors that can be concomitantly found in the CALMS model. Conversely, a large cohort study that aimed to assess the social anxiety, stuttering severity, and speech dissatisfaction of adolescents who stutter showed that they also need cognitive, affective, and social support [16].

Hence, we deemed that adults who stutter should receive CBT as well as speech therapy in their regular treatment. There were reports that the effect of CBT on stuttering was examined by RCT[17,18]. Thirty adults with chronic stuttering were randomly allocated to receive either speech therapy following a CBT treatment for social anxiety or speech therapy alone. The CBT treatment was associated with significant improvements in psychological functioning but did not improve fluency; the participants' subjective evaluation was not examined in this study. Thirty-two adults seeking treatment for stuttering were randomly allocated to receive either speech therapy following an online CBT or speech therapy alone. The online CBT treatment added clinically significant improvement to quality of life[17].

The present pilot study aimed to examine the effectiveness of guided self-help CBT among adults who stutter and have SAD symptoms when it is applied concomitantly to speech therapy.

Ⅱ. Methods

Participants

This study was conducted on patients who came to our hospital complaining of stuttering symptoms at the Department of Otolaryngology at Teikyo University Chiba Medical Center. Inclusion criteria were being aged 12-65 years; meeting the criteria of childhood-onset fluency disorder according to the DSM-5 after being diagnosed with stuttering by an otolaryngologist; and providing written informed consent.

Exclusion criteria were severe psychiatric disorders such as severe schizophrenia spectrum disorders, severe bipolar disorder, increased risk of suicide when enrolling in the study, and substance abuse. Participants diagnosed with common mental disorders (e.g., major

depressive disorder and SAD) by the Mini-International Neuropsychiatric Interview (MINI)[19,20]were eligible for inclusion. Participants could choose to participate in the CBT or control groups. This pilot study was designed as a two-arm, non-randomized controlled trial and performed according to CONSORT guidelines [21].The study was approved by the Ethics Committees of Teikyo University (Teirin17-167) and registered in the University Hospital Medical Information Network Clinical Trials Registry (UMIN000031916). All participants provided written informed consent prior to participation.

Intervention

Regarding usual care, the control group received 20-minute speech therapy sessions once every three or four weeks for four months, for four total sessions of usual care. The first author, as a licensed speech therapist, provided the usual care sessions; the method utilized was fluency shaping (e.g., learning about articulatory movements and practicing vowel production by soft contact of articulators, short sentences, and words used in the workplace). Table 1 shows the protocol for the control group.

Regarding CBT treatment, the CBT group received 60-minute individual CBT sessions every one to two weeks. See Table 2 for the cognitive behavioral therapy (CBT) group protocol.

Based on the Clark and Wells model[12,13]and using a Japanese self-help workbook to treat SAD symptoms[22], the first author, who had completed a CBT training course (Chiba Improving Access to Psychological Therapies project: Chiba-IAPT; [23]) prior to this study and who received individual supervision by the last author, provided participants with guided self-help CBT sessions.

CBT consisted of 14 steps. Two to three steps were done in one session. Participants' homework was to perform each step's tasks before the next treatment day and to read the next step's corresponding workbook chapter as preparation.

The CBT group also received four 20-minute speech therapy sessions as usual care.

Outcome measurements

Our primary clinical outcome measure was the visual analog scale (VAS), which we used to assess the subjective severity of stuttering. The VAS is one of the most widely used instruments in otolaryngology[24], measuring purely subjectively perceptible symptoms. The VAS indicated current stuttering severity on a 10-cm straight line, where "0" is "normal" and "100" is "most severe."

Eight tools were used for the secondary evaluation. First, as all participants were Japanese, we used the Japanese version of the Overall Assessment of the Speaker's Experience of Stuttering (OASES)[25,26]. The tool comprises four subscales and 55 items: Section I, general information (11 items); Section II, reactions to stuttering (15 items); Section III, communication in daily situations (14 items); and Section IV, quality of life (15 items). Each item is scored on a scale from 1 to 5. In section 1, 1 is "always" and 5 is "not at all." In Session 2, 1 is "I don't feel at all" and 5 is "I always feel." In Session 3, 1 is "not difficult at all" and 5 is "extremely difficult." A is the total score for each item, and B is the number of valid responses times 5. The value of A / B x 100 is called the impact score. The impact score ranges between 20-100, and the scores of 20.0-29.9, 30.0-44.9, 45.0-59.9, 60.0-74.9, and

Table 1 Protocol for control group

Session No.	Contents	Times	Details
Session 1	Speech therapy	First time	Patients learn about articulatory movements.
Session 2	Speech therapy	Second time	Patients practice vowel production by soft contact of articulators.
	Midterm evaluation (intermediate)		
Session 3	Speech therapy	Third time	Patients practice short sentences.
Session 4	Speech therapy	Fourth time	Patients practice vocabulary used in the workplace.
	Final evaluation (post)		

Table 2 Protocol for cognitive behavioral therapy（CBT）group

Session No.	Contents	Times		Details
Session 1	CBT	Step 1	Cognitions related to social anxiety	Patients formulate social situations, cognitions, anxiety, and physical reactions.
		Step 2	Safety behaviors	Patients identify and drop dysfunctional safety behaviors.
	Speech therapy	First time		Patients learn about articulatory movements.
Session 2	CBT	Step 3	Attention-shift training	Patients learn how to shift self-focused attention flexibly to the outside.
		Step 4	Behavioral and attentional changes	Patients drop safety behaviors and shift their attention to the outside at the same time.
		Step 5	Video feedback	Patients observe themselves objectively by watching self-taken video.
Session 3	CBT	Step 6	Behavioral experiment 1	Patients differentiate actual results from predicted ones through a real experiment.
		Step 7	Behavioral experiment 2	
	Speech therapy	Second time		Patients practice vowel production by soft contact of articulators.
Session 4	CBT	Step 8	Surveys about other people's thoughts	Patients balance their cognitions to conduct surveys of other people's opinions
		Step 9	Stopping repetitive thoughts	Patients learn how to stop ruminating and worrying before and after social situations.
		Step 10	Imagery rescripting of memories	Patients rescript negative images and traumatic memories of social situations.
Session 5	CBT	Step 11	Schema work	Patients learn to have a different perspective of their schema-level beliefs.
		Step 12	Relapse prevention	Patients relearn how to cope with stressful social situations from the previous CBT steps.
	Midterm evaluation（intermediate）			
Session 6	CBT	Step 13	Assertive communication	Patients learn how to use assertive communication.
		Step 14	Problem-solving	Patients learn six problem-solving steps to encourage solution-focused thinking.
	Speech therapy	Third time		Patients practice short sentences.
Session 7	CBT	Review and summarizing		Patients review and summarize all CBT sessions
	Speech therapy	Fourth time		Patients practice vocabulary used in the workplace.
	Final evaluation（post）			

75.0-100 represent mild, mild to moderate, moderate, moderate to severe, and severe impact of stuttering on quality of life, respectively.

To assess speech fluency, we used a tool developed by Ozawa et al.[27]in which we asked participants to perform an audio-recorded three-minute speech. The audio data were analyzed by the first author by counting the number of blocks, prolongations, and repetitions that could be considered stuttering, which was divided by the total number of uttered phrases times 100 to calculate the appearance rate of the core stuttering symptoms. Speech fluency scores were: 0 to fewer than 3（normal range）, 3 to fewer than 5（very mild）, 5 to fewer than 12（mild）, 12 to fewer than 37（moderate）, and 37 to fewer than 71（severe）. There is no upper limit to this score, and 71 or above is defined as the most severe.

To assess the degree of social anxiety, we used the Liebowitz Social Anxiety Scale（LSAS）[28,29]. The tool comprises 24 items. Each item is scored on a scale from 0 to 3. Based on previous research, a score of 60

or more indicates the participant had SAD[30].

To assess social phobia, we used the Japanese version of the Social Phobia Inventory (SPIN)[31], which comprises 17 items. Scores range from 0 (not applicable at all) to 4 (very true). Its cutoff point is 22 and above in Japanese clinical settings; the higher the score, the more severe the social anxiety[32].

To assess social anxiety of negative evaluations from others, we used the Japanese version of the Short Fear of Negative Evaluation scale (SFNE)[33,34]. The tool comprises 12 items. Each item is scored on a scale from 1 (not applicable at all) to 5 (very true). Higher scores indicate stronger fear of negative ratings.

To assess depression, we used the Patient Health Questionnaire-9 (PHQ-9)[35,36]. The tool comprises nine items. Each item is scored on a scale from 0 (not at all) to 3 (almost every day). The scores correspond to the following: 1-4 points (minor depression), 5-9 points (mild), 10-14 points (moderate), 15-19 points (moderate to severe), and 20-27 points (severe).

To assess generalized anxiety disorder, we used the Generalized Anxiety Disorder-7 (GAD-7)[37,38]. The tool comprises seven items. Each item is scored on a scale from 0 (not at all) to 3 (almost every day). Scores are as follows: 5-9 points (mild), 10-14 points (moderate), and 15-21 points (severe).

To assess health-related quality of life, we used the EuroQol 5-dimension 5-levels (EQ-5D-5L)[39,40]. The tool consists of five subscales (motion, personal control, daily activities, pain/discomfort, anxiety/block) with the following response options (no problems; slight problems; moderate problems; severe problems; extreme problems). The maximum quality of life value of 11111, which represents perfect health, is defined as 1.000, and the minimum of 55555 is defined as -0.025.

All participants were evaluated at three time points: at the start of the interventions (week 0), in the middle of the interventions (during weeks 5-10), and in the final intervention (during weeks 7-14).

Statistical analyses

Data analysis was performed through F-tests and t-tests (paired, unpaired) and Wilcoxon signed-rank test and Mann-Whitney U test using IBM SPSS statistics 22 (IBM Corp., Armonk, NY, USA). Differences before and after intervention in each group are compared using paired t-test and Wilcoxon signed rank test, and the results are shown in the text. Differences between the two groups were compared using the unpaired t-test and the Mann-Whitney U test. Since the results were similar, the unpaired t-test results are shown in the table, and the Mann-Whitney U test results are shown in the text. The differences in the mean scores between time (pre vs. intermediate vs. post) and group (CBT group vs. control group) were assessed using two-way analysis of variance ANOVA (two-way ANOVA) with post hoc Bonferroni test. All *P* values were two-sided; a *p* value of $<.05$ was considered statistically significant. The effect size was calculated using Cohen's d[41], calculated as the difference between both groups' means divided by their pooled standard deviation; a value of >0.20 was deemed a small effect, >0.50 a medium effect, and >0.80 a large effect size.

Ⅲ. Results

Recruitment

Figure 1 shows participants' flow diagram. In total, 36 patients who stuttered visited the otolaryngologist in the relevant hospital during the study. Among these, 18 were under 12 years old, and three patients declined to participate; 21 patients were therefore excluded, which gave us a final sample of 15 participants.

Among our final sample, 10 participants chose the CBT arm, and five chose the control. Nonetheless, in the CBT group, three participants discontinued participation during the study: one started working for a new company after the first session; one started job hunting after the second session; and we lost contact with one after the second session. Hence, their data was excluded from the analyses because they attended lower than 30% of the seven CBT sessions. Finally, seven CBT participants and five control participants who received more than 30% of the total CBT sessions had their collected data analyzed.

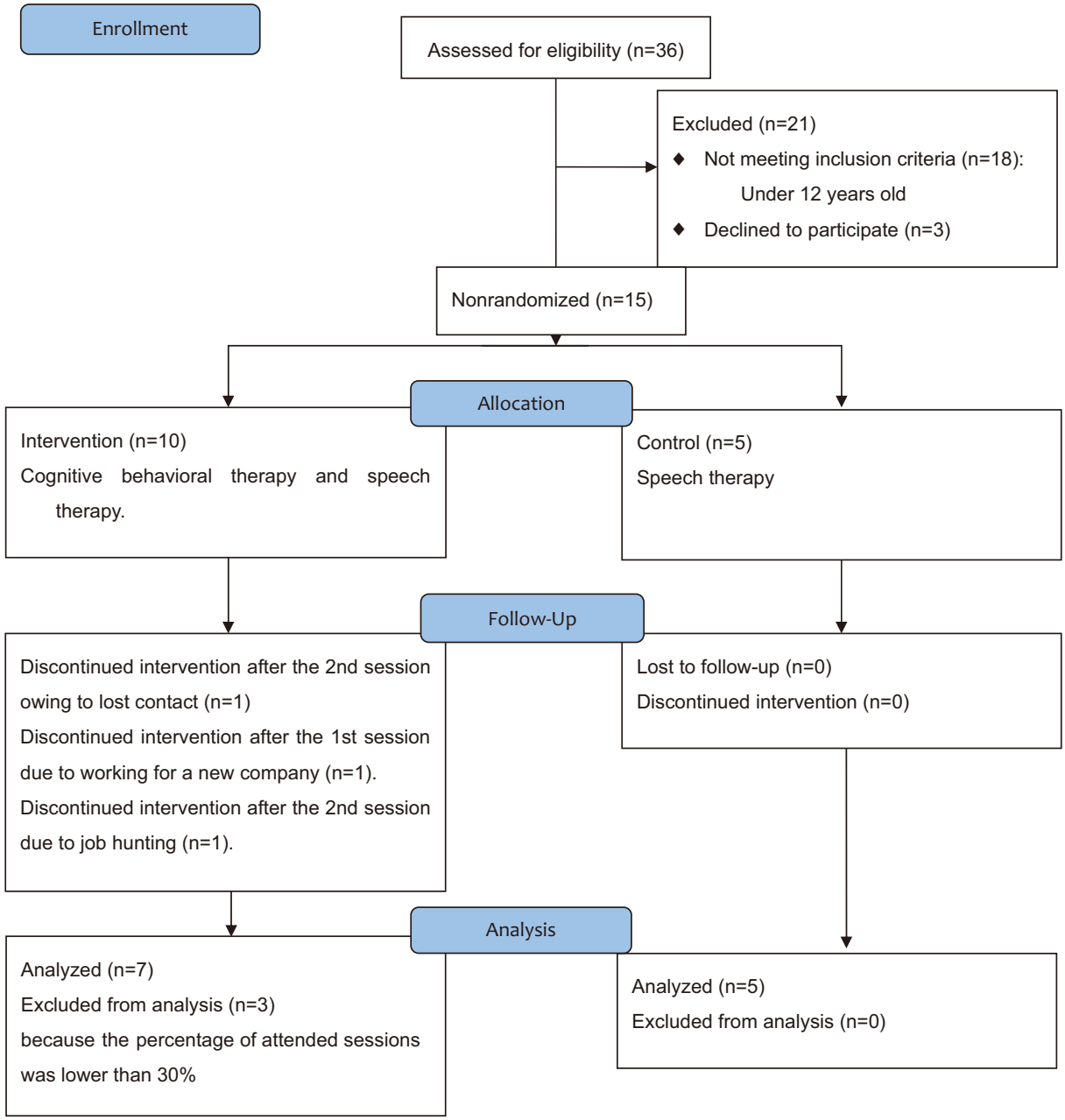

Fig. 1 CONSORT 2010 flow diagram. Of the 36 patients diagnosed with stuttering, 18 children under 12 years of age and 3 patients who did not consent to participate in the study were excluded. Fifteen people were asked to choose between the intervention (CBT) group and the control group. There were 10 patients in the intervention group and 5 patients in the control group. During follow-up, 3 patients in the intervention group withdrew, and the results of 7 patients in the intervention group and 5 patients in the control group were analyzed.

Demographics and clinical characteristics

Table 3 summarizes participants' clinical characteristics at baseline.

In terms of percentage, while 85.7% of the patients (six out of seven) in the CBT group had SAD, 60.0% of the patients (three out of five) in the control group had SAD.

There were no significant differences in the scores for the VAS, OASES, fluency of speech, LSAS, SPIN, PHQ-9, GAD-7, and EQ-5D-5L measures between the two groups. The mean VAS scores at baseline were 57.6 in the CBT group and 53.2 in the control group, meaning both the CBT and control groups were subjectively similar in severity. For the OASES at baseline, mean scores ranged between 67.7 and 71.8 in both groups, suggesting moderate to severe subjective

Table 3 Characteristics of participants at the baseline

Characteristic	CBT (n = 7)	Control (n = 5)	*P* value (unpaired t-test)
Age (years), mean (SD)	26.9 (8.8)	29.8 (11.2)	0.62
Female n (%)	2 (28.6)	2 (40.0)	
Diagnosis n (%)			
Stuttering only	0	1 (20.0)	
Stuttering, social anxiety disorder	3 (42.9)	0	
Stuttering, social anxiety disorder, agoraphobia, bulimia nervosa	1 (14.3)	0	
Stuttering, social anxiety disorder, agoraphobia, panic disorder	1 (14.3)	0	
Stuttering, social anxiety disorder, agoraphobia, major depressive disorder	1 (14.3)	1 (20.0)	
Stuttering, agoraphobia	1 (14.3)	0	
Stuttering, social anxiety disorder, agoraphobia, alcohol abuse	0	1 (20.0)	
Stuttering, social anxiety disorder, agoraphobia, alcohol abuse, major depressive disorder	0	1 (20.0)	
Stuttering, agoraphobia, alcohol dependence, major depressive disorder	0	1 (20.0)	
Total number of stuttering with social anxiety disorder (%)	6 (85.7)	3 (60.0)	
Medication n (%)			
No medication	4 (57.1)	4 (80.0)	
Sertraline	1 (14.3)	0	
Paroxetine, Alprazolam	1 (14.3)	0	
Escitalopram	1 (14.3)	0	
Ethyl Loflazepate	0	1 (20.0)	
Initial evaluation (pre) mean (SD)			
VAS	57.6 (26.1)	53.2 (32.6)	0.80
OASES	67.7 (13.4)	71.8 (16.1)	0.64
Fluency of speech	7.3 (5.9)	10.9 (7.5)	0.37
LSAS	60.7 (21.6)	64.6 (44.5)	0.84
SPIN	37.0 (11.7)	36.0 (11.0)	0.88
SFNE	41.0 (3.4)	34.4 (4.4)	0.01
PHQ-9	8.9 (6.3)	13.6 (7.9)	0.27
GAD-7	9.4 (5.3)	9.0 (4.9)	0.89
EQ-5D-5L	0.800 (0.1)	0.711 (0.2)	0.52

CBT: cognitive behavioral therapy; VAS: Visual analog scale; OASES: Overall Assessment of the Speaker's Experience of Stuttering; LSAS: Liebowitz Social Anxiety Scale; SPIN: Social Phobia Inventory; SFNE: Short Fear of Negative Evaluation scale; PHQ-9: Patient Health Questionnaire-9; GAD-7: Generalized Anxiety Disorder-7; EQ-5D-5L: EuroQol 5-dimension 5-level

distress regarding stuttering behavior. For fluency of speech at baseline, mean scores ranged from 5 to fewer than 12 in both groups, suggesting mild stuttering from the observers' viewpoint. For LSAS at baseline, the mean scores were 60 or more in both groups, suggesting the presence of SAD symptoms. For the SPIN at baseline, the mean scores were over 22 in both groups, also suggesting the presence of SAD symptoms.

The only scale with significant differences was the SFNE, with an average score of 41.0 in the CBT group and 34.4 in the control group. This means that the average score at baseline was significantly higher in the CBT group compared to the control group ($p < 0.01$), indicating that the CBT group was very anxious about negative ratings from others.

Primary outcome

Figure 2 shows the results for each evaluation. The solid line shows the CBT group and the dashed line shows the mean and standard deviation of the control group pre, intermediate, and post. The results of the two-way ANOVA showed that there was no interaction

between time and group (F (2,9) = 3.96, n.s.). Table 4 show the results of the primary outcome measure by time period and group. Regarding the VAS, the mean change in the final evaluation compared to the baseline evaluation was -16.14 (95% CI[-39.08, +6.80]) and +10.00 (95% CI[-8.54, +28.54]) for the CBT and control groups, respectively. The differences between groups were non-significant, but the decrease was greater in the CBT group (-26.14, $p = 0.06$), and the effect size was large (Cohen's d = 1.22). In addition, the results of difference between pre- and post- intervention in each group using the paired t-test were not significant ($p = 0.14$ in the CBT group and $p = 0.21$ in the control group). The Wilcoxon signed-rank test, treated VAS as non-parametric data, also showed no significant differences ($p = 0.13$ in the CBT group and $p = 0.14$ in the control group).

Secondary outcomes

Figure 2 and Table 4 show participants' detail scores in the secondary outcome measures. The results of two-way ANOVA were as follows. Regarding OASES, there was no interaction between time and group (F (2,9) = 2.49, n.s.). Regarding fluency of speech, there was no interaction between time and group (F (2,9) = 0.37, n.s.). Regarding LSAS-J, there was no interaction between time and group (F (2,9) = 1.07, n.s.). Regarding SPIN, there was no interaction between time and group (F (2,9) = 0.42, n.s.). Regarding SFNE, there was an significant interaction between time and group (F (2,9) = 5.35, $p = 0.03$.), and post-hoc Bonferroni test showed a significant deterioration between the intermediate and post evaluations in the control group ($p = 0.04$). Regarding PHQ-9, there was no interaction between time and group (F (2,9) = 0.20, n.s.). Regarding GAD-7, there was no interaction between time and group (F (2,9) = 2.91, n.s.). Regarding EQ5D5L, there was no interaction between time and group (F (2,9) = 0.30, n.s.).

Regarding the OASES, the mean change in the final evaluation compared to the baseline evaluation was -11.12 (95% CI[-24.80, +2.56]) and -2.48 (95% CI[-9.02, +4.06]) in the CBT and control groups, respectively. The difference between groups was non-significant (-8.64; $p = 0.19$), but the effect size was medium (Cohen's d = 0.72).

Regarding fluency of speech, the mean change in the final evaluation compared to the baseline evaluation was +0.37 (95% CI[-6.61, +7.36]) and -1.50 (95% CI[-7.00, +4.00]) in the CBT and control groups, respectively. The difference between groups was non-significant (+1.87; $p = 0.63$), and the effect size was small (Cohen's d = 0.29).

Regarding the LSAS, the mean change in the final evaluation compared to the baseline evaluation was -13.71 (95% CI[-41.44, +14.01]) and +6.80 (95%CI [-11.83, +25.43]) in the CBT and control groups, respectively. The difference between groups was non-significant, (-20.51; $p = 0.19$), but the effect size was large (Cohen's d = 0.82).

Regarding the SPIN, the mean change in the final evaluation compared to the baseline evaluation was -9.29 (95% CI[-24.79, +6.21]) and -1.20 (95% CI [-17.30, +14.90]) in the CBT and control groups, respectively. The difference between groups was non-significant (-8.09; $p = 0.39$), but the effect size was medium (Cohen's d = 0.54).

Regarding the SFNE score, the mean change in the final evaluation compared to the baseline evaluation was -5.14 (95% CI[-10.93, +0.64]) in the CBT group and +3.60 (95% CI[+0.61, +6.59]) in the control group. The improvement in SFNE in the CBT group was significantly larger than that in the control group (difference between two groups = -8.74; unpaired t test, $p < .01$, Mann-Whitney U-test: $p < .05$), and the effect size was large (Cohen's d = 1.70).

The SFNE score in the control group showed a significant deterioration between pre- and post-intervention using the paired t-test ($p = 0.03$). This result was also significant in the Wilcoxon signed rank test ($p = 0.04$). The SFNE score in the CBT group showed a non-significant change between pre- and post-intervention using the paired t-test ($p = 0.07$). This result was not significant in the Wilcoxon signed rank test ($p = 0.06$).

The mean SFNE score in the CBT group was

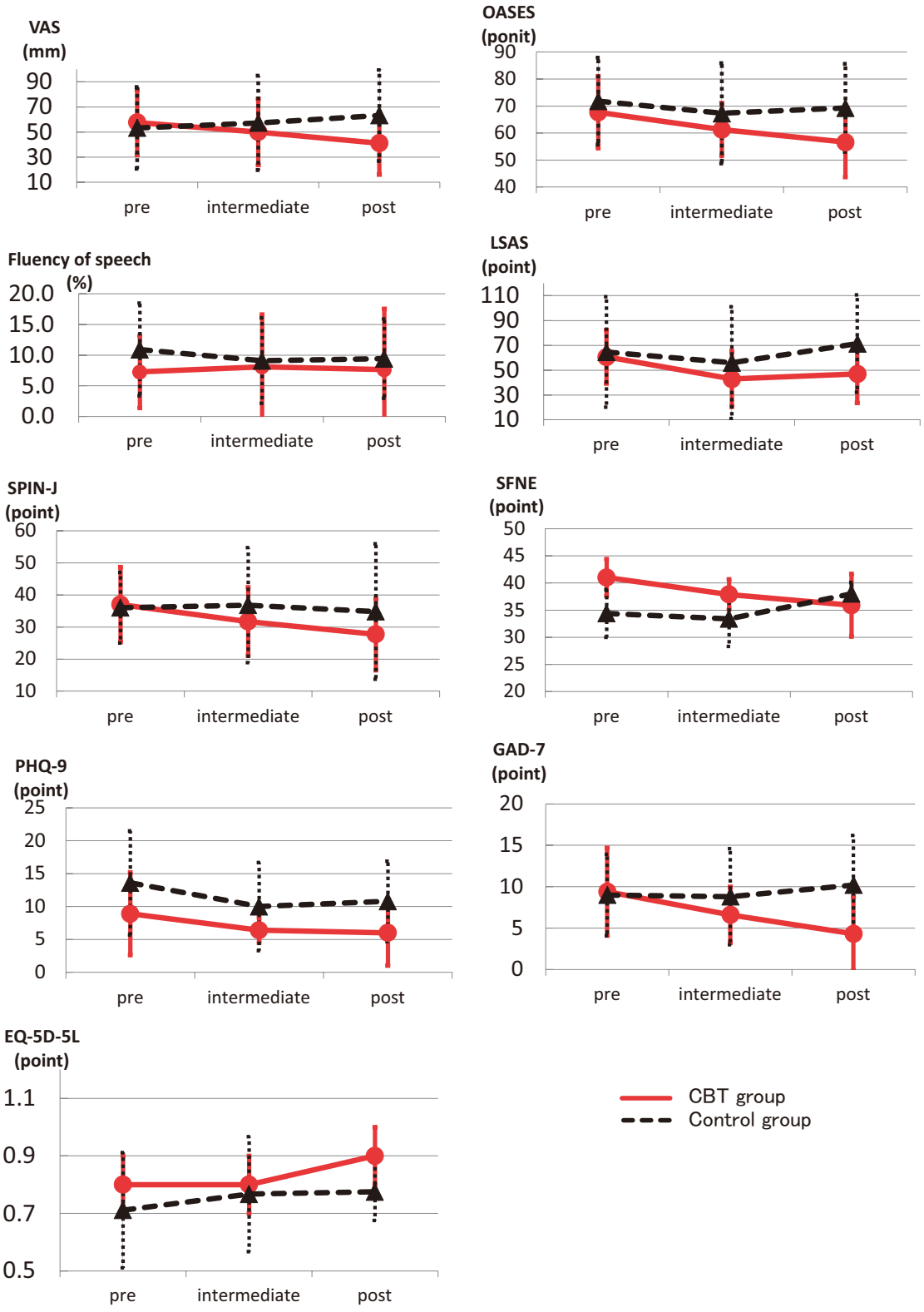

Fig. 2 Graphs showing outcomes at each assessment. CBT: cognitive behavioral therapy; VAS: Visual analog scale; OASES: Overall Assessment of the Speaker's Experience of Stuttering; LSAS: Liebowitz Social Anxiety Scale; SPIN: Social Phobia Inventory; SFNE: Short Fear of Negative Evaluation scale; PHQ-9: Patient Health Questionnaire-9; GAD-7: Generalized Anxiety Disorder-7; EQ-5D-5L: EuroQol 5-dimension 5-level. This is the result of the mean ± standard deviation of the intervention group (CBT group) and control group (control group) at the baseline (pre), intermediate evaluation (intermediate), and final evaluation (post) of the primary and secondary evaluations. CBT group is a red straight line, control group is a black dashed line, ● ▲ is the mean value, and shadows are ± standard deviation.

Table 4 Changes in outcomes

Changes from baseline	CBT (n = 7)	95%CI	Control (n = 5)	95% CI	Intergroup difference	*P*-value (unpaired t-test)	effect size (d)
Primary outcome							
VAS	-16.14	-39.08 to 6.80	10.00	-8.54 to 28.54	-26.14	0.06	1.22
Secondary outcomes							
OASES	-11.12	-24.80 to 2.56	-2.48	-9.02 to 4.06	-8.64	0.19	0.72
Fluency of speech	0.37	-6.61 to 7.36	-1.50	-7.00 to 4.00	1.87	0.63	0.29
LSAS	-13.71	-41.44 to 14.01	6.80	-11.83 to 25.43	-20.51	0.19	0.82
SPIN	-9.29	-24.79 to 6.21	-1.20	-17.30 to 14.90	-8.09	0.39	0.54
SFNE	-5.14	-10.93 to 0.64	3.60	0.61 to 6.59	-8.74	0.01	1.70
PHQ-9	-2.86	-11.46 to 5.74	-2.80	-13.17 to 7.57	-0.06	0.99	0.01
GAD-7	-5.14	-11.80 to 1.51	1.20	-4.31 to 6.71	-6.34	0.11	1.01
EQ-5D-5L	0.101	-0.04 to 0.24	0.064	-0.08 to 0.21	0.037	0.65	0.27

CBT: cognitive behavioral therapy; VAS: Visual analog scale; OASES: Overall Assessment of the Speaker's Experience of Stuttering; LSAS: Liebowitz Social Anxiety Scale; SPIN: Social Phobia Inventory; SFNE: Short Fear of Negative Evaluation scale; PHQ-9: Patient Health Questionnaire-9; GAD-7: Generalized Anxiety Disorder-7; EQ-5D-5L: EuroQol 5-dimension 5-level

significantly higher than that in the control group using the unpaired t-test at the baseline (pre) ($p = 0.01$). On the contrary, the mean SFNE score in the CBT group was significantly lower than that in the control group using the unpaired t-test at the final evaluation (post) ($p = 0.01$). The results using the Mann-Whitney U test, treated SFNE as non-parametric data, also showed a significant difference at the baseline (pre) ($p = 0.01$), but non-significant difference at the final evaluation (post) SFNE score ($p = 0.81$).

Regarding the PHQ-9, the mean change in the final evaluation compared to the baseline evaluation was -2.86 (95% CI[-11.46, +5.74]) and -2.80 (95% CI [-13.17, +7.57]) in the CBT and control groups, respectively. The difference between groups was non-significant (-0.06; $p = 0.99$), and the effect size was too low (Cohen's d = 0.01).

Regarding the GAD-7, the mean change in the final evaluation compared to the baseline evaluation was -5.14 (95% CI[-11.80, +1.51]) and +1.20 (95% CI[-4.31, +6.71]) in the CBT and control groups, respectively. The difference between groups was non-significant (-6.34; $p = 0.11$), but the effect size was large (Cohen's d = 1.01).

Regarding the EQ-5D-5L, the mean change in the final evaluation compared to the baseline evaluation was +0.101 (95% CI[-0.04, +0.24]) and +0.064 (95%CI[-0.08, +0.21]) in the CBT and control groups, respectively. The difference between groups was non-significant (+0.037; $p = 0.65$), and the effect size was small (Cohen's d = 0.27).

Table 5 shows changes in the OASES subscales. Regarding the total impact score, the average change (SD) for the final evaluation compared to the baseline was -11.1 (14.8) and -2.5 (5.3) in the CBT and control groups, respectively. The difference between groups was non-significant, the decrease was greater in the CBT group (-8.6; $p = 0.19$), and the effect size was medium (Cohen's d = 0.72).

Regarding Section Ⅰ (general information), the average change (SD) for the final versus baseline evaluation was -8.1 (13.0) and -5.5 (6.2) in the CBT and control groups, respectively. The difference between groups was non-significant, the decrease was greater in the CBT group (-2.6; $p = 0.70$), and the effect size was small (Cohen's d = 0.24).

Regarding Section Ⅱ (reactions to stuttering), the average change (SD) at the final evaluation compared to the baseline evaluation was -11.2 (24.4) and -1.8 (7.8) in the CBT and control groups, respectively. The difference between groups was non-significant, the decrease was greater in the CBT group (-9.4; $p = 0.37$),

Table 5 Changes in OASES subscales

Changes from baseline	CBT (n = 7)	standard deviation	Control (n = 5)	standard deviation	Intergroup difference	*P*-value (unpaired t-test)	effect size (d)
Total Impact Score	-11.1	14.8	-2.5	5.3	-8.6	0.19	0.72
Section I: General Information	-8.1	13.0	-5.5	6.2	-2.6	0.70	0.24
Section II: Reactions to Stuttering	-11.2	24.4	-1.8	7.8	-9.4	0.37	0.48
Section III: Communication in Daily Situations	-6.1	11.7	3.3	8.3	-9.4	0.16	0.90
Section IV: Quality of Life	-15.6	20.4	-4.3	6.9	-11.3	0.22	0.69

CBT: cognitive behavioral therapy; OASES: Overall Assessment of the Speaker's Experience of Stuttering

and the effect size was small (Cohen's d = 0.48).

Regarding Section Ⅲ (communication in daily situations), the average change (SD) at the final evaluation compared to the baseline was -6.1 (11.7) and 3.3 (8.3) in the CBT and control groups, respectively. The difference between groups was non-significant, the decrease was greater in the CBT group (-9.4; $p = 0.16$), and the effect size was large (Cohen's d = 0.90).

Regarding Section Ⅳ (quality of life), the average change (SD) at the final evaluation compared to the baseline evaluation was -15.6 (20.4) and -4.3 (6.9) in the CBT and control groups, respectively. The difference between groups was non-significant, the decrease was greater in the CBT group (-11.3; $p = 0.22$), and the effect size was medium (Cohen's d = 0.69).

Ⅳ. Discussion

This study investigated the effectiveness of guided self-help CBT in adults with stuttering in an intervention group that combined CBT with speech therapy and a speech therapy-only control group. Prior studies have examined CBT's effects on people who stutter using randomized controlled trials[17,18], but have not assessed participants' subjective stuttering severity. Therefore, the current study is the first to use guided self-help CBT for SAD based on the Clark and Wells model to decrease subjective stuttering severity. The VAS, OASES, fluency of speech, LSAS, SPIN, SFNE, PHQ-9, GAD-7, and EQ-5D-5 were evaluated pre-, mid-, and post treatment. Although there was almost no statistically significant difference between the CBT and control group, results of the VAS, OASES, LSAS, SPIN, SFNE, and GAD-7 showed the intervention had a medium to large effect size on stuttering.

As the secondary outcome, CBT group showed better improvement of the SFNE score compared with control group. This result suggested that CBT intervention may be effective in reducing fear of negative evaluation for stutters.

The study showed that subjective severity of stuttering and VAS and OASES scores tended to improve in the CBT group, particularly for Section III of the OASES, which includes questions concerning how difficult is it for respondents to talk in front of a large group, continue to speak regardless of how the listener responds, and order food in a restaurant. As these situations reflect subjective distress in daily communication, improved scores mean the person who stutters is more comfortable with daily communication. In other words, the results suggest CBT can improve the quality of life of people who stutter.

Regarding objective speech fluency before and after treatment, we found no apparent difference; the severity of stuttering in both groups remained mild before and after treatment. Interestingly, while fluency of speech scores were midrange, subjective stuttering severity measured using VAS scores was high before treatment.

SAD is often reported to be associated with major depressive disorder[42,43]. In treating patients with both stuttering and SAD, psychiatrists and otolaryngologists need to cooperate. Otolaryngologists tend to note SAD symptoms among these patients, and they should then recommend the patient consult a psychiatrist who can help them manage SAD symptoms; however, many patients who visit doctors with

complaints about stuttering tend to refuse consultation with a psychiatrist because their problem relates to stuttering behavior, not psychological issues. Initially, in our study, most patients reported stuttering symptoms but did not report SAD symptoms until asked by an otolaryngologist. Therefore, the otolaryngologist should perform a medical examination to assess whether the patient who stutters has SAD.

As previously mentioned, one participant dropped out of the study because of a new job. Although the participant wanted to continue CBT and speech therapy, it was unclear if CBT and speech therapy were available together at the same facility, and the participant mentioned the difficulty for working adults to visit two departments（i.e., otolaryngology and psychiatry）because of time constraints. In this study, the combination of CBT and speech therapy showed a tendency for improvement in the studied sample, although it either was not statistically significant. The CBT group had lower LSAS scores than the control group, suggesting that social anxiety symptoms were reduced after the interventions. Therefore, if speech therapists have the option and skills to promote a CBT intervention together with speech therapy, our results corroborate the possibility that this conjunction can provide more effective support for adults who stutter. The self-guided CBT used in this study is structured in a way that is easy for both speech therapists and people who stutter to follow. However, speech therapists should be supervised by psychiatrists.

Several limitations of this study should be acknowledged, including that it was a nonrandomized controlled trial, had a small sample size, and did not include long-term follow-up. In future studies, randomized controlled trials with larger samples and follow-up for a longer period of time should be conducted to confirm the findings that CBT may improve subjective severity of stuttering.

Contributors

F.O. conceived, designed, conducted, analyzed, and interpreted the study, with the help of A.N., R.H., M.S., and E.S. All authors discussed the data and results and critically reviewed the manuscript. All authors approved the final version of the manuscript.

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Conflict of interest

The authors declare that they have no conflicts of interest, either financial or non-financial, with the contents of this article.

E.S. is a member of the Editorial Board of the Chiba Medical Journal.

Ethical approval

The study was approved by the Ethics Committees of Teikyo University（Teirin17-167）and registered in the University Hospital Medical Information Network Clinical Trials Registry（UMIN000031916）. All participants provided written informed consent prior to participation.

Data availability

The data that support the findings of this study are available from the corresponding author, F. O., upon reasonable request.

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References

1）American Psychiatric Association.（2013）Diagnostic and Statistical Manual of Mental Disorders 5th Edition. Arlington: American Psychiatric Association Publising.

2）Reilly S, Onslow M, Packman A, Cini E, Conway L, Ukoumunne OC, Bavin EL, Prior M, Eadie P, Block S, Wake M.（2013）Natural history of stuttering to 4 years of age: a prospective community-based study. Pediatrics 132, 460-7.

3）Yairi E, Ambrose N.（2013）Epidemiology of stuttering: 21st century advances. J Fluency Disord 38, 66-87.

4）Howell P, Davis S.（2011）Predicting persistence of and recovery from stuttering by the teenage years based on information gathered at age 8 years. J Dev Behav Pediatr 32, 196-205.

5）Blood GW, Blood IM.（2004）Bullying in adolescents who Stutter: communicative competence and self-esteem. Contemp Issues Commun Sci Disord 31, 69-79.

6）Iverach L, Rapee RM.（2014）Social anxiety disorder and stuttering: current status and future directions. J Fluency Disord 40, 69-82.

7）Blood GW, Blood IM.（2016）Long-term consequences of childhood bullying in adults who stutter: social anxiety, fear of negative evaluation, self-esteem, and satisfaction with life. J Fluency Disord 50, 72-84.

8）Stein DJ, Lim CCW, Roest AM, de Jonge P, Aguilar-Gaxiola S, Al-Hamzawi A, et al. WHO World Mental Health Survey Collaborators.（2017）The cross-national epidemiology of social anxiety disorder: data from the World Mental Health Survey Initiative. BMC Med 15, 143.

9）Blumgart E, Tran Y, Craig A.（2010）Social anxiety disorder in adults who stutter. Depress Anxiety 27, 687-92.

10）Mayo-Wilson E, Dias S, Mavranezouli I, Kew K, Clark DM, Ades AE, Pilling S.（2014）Psychological and pharmacological interventions for social anxiety disorder in adults: a systematic review and network meta-analysis. Lancet Psychiatry 1, 368-76.

11）Clark DM, Ehlers A, McManus F, Hackmann A, Fennell M, Campbell H, Flower T, Davenport C, Louis B.（2003）Cognitive therapy versus fluoxetine in generalized social phobia: a randomized placebo-controlled trial. J Consult Clin Psychol 71, 1058-67.

12）Yoshinaga N, Matsuki S, Niitsu T, Sato Y, Tanaka M, Ibuki H, Takanashi R, Ohshiro K, Ohshima F, Asano K, Kobori O, Yoshimura K, Hirano Y, Sawaguchi K, Koshizaka M, Hanaoka H, Nakagawa A, Nakazato M, Iyo M, Shimizu E.（2016）Cognitive behavioral therapy for patients with social anxiety disorder who remain symptomatic following antidepressant treatment: a randomized, assessor-blinded, controlled trial. Psychother Psychosom 85, 208-17.

13）Yoshinaga N, Kubota K, Yoshimura K, Takanashi R, Ishida Y, Iyo M, Fukuda T, Shimizu E.（2019）Long-term effectiveness of cognitive therapy for refractory social anxiety disorder: one-year follow-up of a randomized controlled trial. Psychother Psychosom 88, 244-6.

14）Healey EC, Trautman LS, Susca M.（2004）Clinical applications of a multidimensional approach for the assessment and treatment of stuttering. Contemporary Issues in Communication Science and Disorders 31, 40-8.

15）Perez HR, Stoeckle JH.（2016）Stuttering: clinical and research update. Can Fam Phys 62, 479-84.

16）Iverach L, Rapee RM, Wong QJJ, Lowe R.（2017）Maintenance of social anxiety in stuttering: a cognitive-behavioral model. Am J Speech Lang Pathol 26, 540-56.

17）Menzies R, O'Brian S, Packman A, Jones M, Helgadóttir FD, Onslow M.（2019）Supplementing stuttering treatment with online cognitive behavior therapy: an experimental trial. J Commun Disord 80, 81-91.

18）Menzies RG, O'Brian S, Onslow M, Packman A, St Clare T, Block S.（2008）An experimental clinical trial of a cognitive-behavior therapy package for chronic stuttering. J Speech Lang Hear Res 51, 1451-64.

19）Otsubo T, Tanaka K, Koda R, Shinoda J, Sano N, Tanaka S, Aoyama H, Miura M, Kamijima K.（2005）Reliability and validity of Japanese version of the mini-international neuropsychiatric interview. Psychiatry Clin Neurosci 59, 517-26.

20）Sheehan DV, Lecrubier Y, Sheehan KH, Amorim P, Janavs J, Weiller E, Hergueta T, Baker R, Dunbar GC.（1998）The mini-international neuropsychiatric interview（M.I.N.I.）: the development and validation of a structured diagnostic psychiatric interview for DSM-IV and ICD-10. J Clin Psychiatry 59（suppl 20）, 22-33.

21）Schulz KF, Altman DG, Moher D.（2010）CONSORT 2010 statement: updated guidelines for reporting parallel group randomised trials. BMJ 340, c332.

22）Shimizu E.（2014）Jibunde Naosu "Shakou Fuan Shougai": Jibunni Jisinga Moteru（Japanese Edition）. Tokyo: houken.

23）Kobori O, Nakazato M, Yoshinaga N, Shiraishi T, Takaoka K, Nakagawa A, Iyo M, Shimizu E.（2014）Transporting cognitive behavioral therapy（CBT）and the improving access to psychological therapies（IAPT）project to Japan: preliminary observations and service evaluation in Chiba. J Men Health Train Educ Pract 9, 155-66.

24）Adamchic I, Langguth B, Hauptmann C, Alexander TP.（2012）Psychometric evaluation of visual analog scale for the assessment of chronic tinnitus. Am J Audiol 21, 215-25.

25）Sakai N, Chu SY, Mori K, Yaruss JS.（2017）The Japanese version of the overall assessment of the speaker's experience of stuttering for adults（OASES-A-J）: translation and psychometric evaluation. J Fluency Disord 51, 50-9.

26）Yaruss JS, Quesal RW.（2006）Overall assessment of the speaker's experience of stuttering（OASES）: documenting multiple outcomes in stuttering treatment. J Fluency Disord 31, 90-115.

27）Ozawa E, Hara Y, Suzuki N, Moriyama H, Ohashi Y, Mochida A, Sakata Y, Sakai N.（2016）Kitsuon Kensahou Dai 2 han Kaisetsu 2nd Edition.（in Japanese）. Tokyo: Gakuensya.
28）Asakura S, Inoue S, Sasaki F, Sasaki Y, Kikagawa N, Inoue T, Denda K, Ito K, Matsubara R, Koyama T.（2002）Reliability and validity of the Japanese version of the Liebowitz social anxiety scale.（in Japanese）. Seishin Igaku（Clinical Psychiatry）44, 1077-84.
29）Liebowitz MR, Gorman JM, Fyer AJ, Klein DF.（1985）Social phobia. review of a neglected anxiety disorder. Archi Gen Psychiatry 42, 729-36.
30）Mennin DS, Fresco DM, Heimberg RG, Schneier FR, Davies SO, Liebowitz MR.（2002）Screening for social anxiety disorder in the clinical setting: using the Liebowitz social anxiety scale. J Anxiety Disord 16, 661-73.
31）Connor KM, Davidson JR, Churchill LE, Sherwood A, Foa E, Weisler RH.（2000）Psychometric properties of the Social Phobia Inventory（SPIN）. new self-rating scale. Br J Psychiatry 176, 379-86.
32）Nagata T, Nakajima T, Teo AR, Yamada H, Yoshimura C.（2013）Psychometric properties of the Japanese version of the social phobia inventory. Psychiatry Clin Neurosci 67, 160-6.
33）Sasagawa S, Kanai Y, Muranaka Y, Suzuki S, Shimada H, Sakano Y.（2004）Development of a short fear of negative evaluation scale for Japanese using item response theory. Japanese Journal of Behavior Therapy 30, 87-98.
34）Watson D, Friend R.（1969）Measurement of social-evaluative anxiety. J Consult Clin Psychol 33, 448-57.
35）Muramatsu K, Miyaoka H, Kamijima K, Muramatsu Y, Yoshida M, Otsubo T, Gejyo F.（2007）The patient health questionnaire, Japanese version: validity according to the mini-international neuropsychiatric interview-plus. Psychol Rep 101, 952-60.
36）Spitzer RL, Kroenke K, Williams JB.（1999）Validation and utility of a self-report version of PRIME-MD: the PHQ primary care study. JAMA 282, 1737-44.
37）Muramatsu K.（2014）An up-to-date letter in the Japanese version of PHQ, PHQ-9, PHQ-15. Niigata Seiryo Daigakudaigakuin Rinshosinrigakukenkyu（Grad School of Niigata Seiryo Univ Clin Psychol Res）7, 35-9.
38）Spitzer RL, Kroenke K, Williams JBW, Löwe B.（2006）A brief measure for assessing generalized anxiety disorder: the GAD-7. Arch Internal Med 166, 1092-7.
39）EuroQol Group.（1990）EuroQol－a new facility for the measurement of health-related quality of life. Health Policy 16, 199-208.
40）Tsuchiya A, Ikeda S, Ikegami N, Nishimura S, Sakai I, Fukuda T, Hamashima C, Hisashige A, Tamura M.（2002）Estimating an EQ-5D population value set: the case of Japan. Health Econ 11, 341-53.
41）Cohen J.（1977）Statistical power analysis for the behavioral sciences 2nd Edition. New York: Psychology Press.
42）Dalrymple KL, Zimmerman M.（2007）Does comorbid social anxiety disorder impact the clinical presentation of principal major depressive disorder? J Affec Disord 100, 241-7.
43）Stein MB, Chavira DA.（1998）Subtypes of social phobia and comorbidity with depression and other anxiety disorders. J Affec Disord 50, S11-S6.