

## The impact of social anxiety disorder on the duration of speech and language therapy at a medical institution for Japanese individuals who stutter

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**Objective:** To investigate the relationship between social anxiety disorder (SAD) tendency and the implementation of speech and language therapy (SLT) in adult stutterers who visit medical institutions.

**Methods:** Eighty-eight adults (average age 26.7 years; 66 men, 22 women) who were diagnosed with stuttering, based on the criteria proposed in the Diagnostic and Statistical Manual of Mental Disorders (DSM-5), and started SLT, were recruited. From the medical records, basic information, information on the duration of SLT, stuttering frequency of reading, and the test data of the Japanese version of the Liebowitz Social Anxiety Scale (LSAS-J) were collected retrospectively. The patients were then divided into 2 groups: those with and those without SAD, and the outcomes at 6 months after the termination of the SLT were compared.

**Results:** The group without SAD contained a significantly higher percentage of individuals who completed training ( $P < 0.01$ ) than did the group with SAD. The LSAS-J total score at the end of the training in the group that completed the training was significantly lower than that at the first visit ( $P < 0.01$ ).

**Conclusion:** We concluded that the presence or absence of SAD tended to influence the implementation of SLT.

**Key words:** stuttering, adult, social anxiety, LSAS-J, speech and language therapy

**Abbreviations:** SAD, social anxiety disorder; DSM, Diagnostic and Statistical Manual of Mental Disorders; SLT, speech and language therapy; LSAS-J, Liebowitz Social Anxiety Scale-Japanese version

### Introduction

Stuttering is a speech fluency disorder whose core symptoms are sound and word repetition, stretching sounds, and blocks of speech.<sup>1</sup> The incidence of stuttering in the general population in the West is about 5%<sup>2</sup>; and in Japan, it is reported to be 1.4% at the over 3-year-old point.<sup>3</sup> The prevalence of stuttering in adults is 0.8%.<sup>4</sup> Stuttering disorders coexist with various neuropsychiatric diseases,<sup>2</sup> of these the one that has received special attention in recent years is social anxiety disorder (SAD).<sup>5</sup>

SAD is a mental illness characterized by fear and

avoidance of social situations such as public conversation, writing, eating, and drinking in public places, and meeting with strangers.<sup>1</sup> Epidemiologically, about 75% of SAD cases occur between the ages of 8 and 15, and the occurrence is similar or slightly higher in women than in men.<sup>1</sup> The lifetime prevalence of SAD in the general public is estimated to be 8%–13%.<sup>6-7</sup> Treatment of SAD usually involves psychopharmaceutical therapy<sup>8-9</sup> with selective serotonin reuptake inhibitors, and cognitive-behavioral therapy,<sup>10-11</sup> alone or in parallel.

In a large cohort study of 102 patients who visited a stuttering treatment facility between the ages of 11 and

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17, about 70% of the patients suffered from neuropsychiatric disorders including SAD.<sup>12</sup> Furthermore, a screening showed that more than 40% of 200 adult stutterers also suffered from SAD.<sup>13</sup> In addition to clinical symptoms, SAD interferes with social life such as schoolwork and employment as a secondary disability, leading to a decline in the quality of life.<sup>14</sup> Therefore, it is considered that the effects of SAD on people who stutter are not small.<sup>15</sup>

Speech and language therapy (SLT) for stuttering can be roughly classified into a direct method that directly affects speech and an indirect method that indirectly affects speech. In particular, one of the direct methods, the fluency shaping approach, has been reported to date with considerable evidence regarding the training effect.<sup>16-18</sup> The fluency shaping approach is a method of establishing a way of speaking that does not cause stuttering inside the training room but generalizes it outside the training room. Speaking techniques that make stuttering less likely include skills, such as slower speech rate, soft voice, light contact with articulatory organs, and smooth expiratory breathing within the expiratory paragraph.

Using the fluency shaping approach, it is possible for stutterers to repeat the successful experience of reducing the stuttering symptoms in the speech treatment period (in a training room). However, it was pointed out that it is difficult to generalize that successful experience in daily life (outside the training room) and maintain a positive behavioral change.<sup>16</sup> As one of the factors, it is considered that the fluency shaping approach cannot sufficiently remove the psychological reactions of stutterers, such as the anxiety and fear of stuttering and the innate avoidance behavior common to many stutterers.<sup>19</sup>

In addition, considering the high rate of coexistence of stuttering with SAD, psychological reactions, such as anxiety, fear, and avoidance behavior may occur not only because of stuttering but also due to the coexisting SAD. Therefore, it is important to consider the severity of SAD in addition to the severity of their stuttering symptoms when using the fluency shaping approach on stutterers with SAD. However, it has not been examined how the SAD tendency of the stutterers affects the duration of SLT using the fluency shaping approach.

The objective of this study was to investigate the relationship between SAD and the necessary duration of SLT in adults who stutter.

## Materials and Methods

### *Patients*

From June 2013 to May 2014, we enrolled 101 patients (18 years of age or older) who visited the Department of Rehabilitation, Kitasato University East Hospital with a stuttering complaint. Thirteen cases undergoing psychopharmaceutical therapy were excluded, and the final number of cases in the present study was 88.

A physician diagnosed stuttering in these 88 cases based on the Diagnostic and Statistical Manual of Mental Disorder (DSM-5) criteria for stuttering, and SLT was started by a speech-language-hearing therapist. The average age of the 88 patients was  $26.7 \pm 7.2$  years (age range 18–50 years); there were 66 men and 22 women. Three of the 88 cases had neurodevelopmental disorders in addition to stuttering (1 with autism spectrum disorder, 1 with localized learning disorder, and 1 with both autism spectrum disorder and attention deficit hyperactivity disorder).

### Methods

From the medical records, patients basic information (age, gender, presence or absence of comorbidities, and medication history, etc.), information on the duration of SLT, stuttering frequency of reading sentences, and inspection data of the Japanese version of the Liebowitz Social Anxiety Scale (LSAS-J)<sup>20</sup> were collected retrospectively. Since 2011, stuttering frequency of reading sentences and inspection data of the LSAS-J have been obtained for all patients with stuttering when first examined.

Next, patients were divided into 2 groups: those with and without SAD, based on the LSAS-J cutoff value, and the outcomes were compared 6 months after the commencement of SLT. Furthermore, the LSAS-J was re-examined at 6 months after the commencement of the therapy, and the scores were compared with those at the first visit. The reasons for the re-examination 6 months after the commencement of SLT were as follows. In treatment covered by the National Japanese Health Insurance, a cerebrovascular rehabilitation fee is calculated for SLT. The standard calculation days for cerebrovascular rehabilitation fees are limited to 180 days or less. Therefore, we evaluated the clinical outcome at 6 months after the commencement of SLT.

This study was approved by the Ethics Committee of Kitasato University School of Medicine (B14-44).

## Inspection data

### *Stuttering frequency when reading aloud*

After a physician diagnoses stuttering, a speech-language-hearing therapist measures the frequency of stuttering in a brief sentence-reading period. The sentences from Aesop's fairy tale "North Wind and the Sun" (52 passages) were used for the reading task. The speech at the time of reading was recorded and saved using the SUGI Speech Analyzer software (Animo, Tokyo). The speech data were analyzed by 3 speech-language-hearing therapists each having more than 10 years of clinical experience with stuttering. Stuttering symptoms were identified visually and aurally by presenting the waveform of speech data on a personal computer screen and then reproducing the speech.<sup>21</sup> The stuttering frequency was obtained from the number of phrases in which the core symptoms of the stuttering appeared (sound and word repetition, stretching, and blocks), divided by the total number of sentences read aloud (52 phrases), multiplied by 100.

### *The Liebowitz Social Anxiety Scale Japanese version (LSAS-J)*

The LSAS-J is composed of 24 items: 13 items related to behavior, and 11 items related to social status; it is a Likert scale that measures the degree of "fear/anxiety" and "avoidance" in 4 stages (0–3). The LSAS total score is rated from 0 to 144, with higher scores indicating higher social anxiety. Fresco et al.<sup>22</sup> observed that the LSAS total score was 13.5 (standard deviation [SD], 12.7) for healthy patients and 73.4 (SD, 23.2) for SAD patients. An LSAS score of 90 or higher is associated with serious problems in daily life.<sup>23</sup> The LSAS is a useful screening tool for SAD.<sup>24</sup> The Japanese version of the LSAS, the LSAS-J, has a high internal consistency ( $\alpha = 0.95$ ), reliability, and sufficient convergent validity.<sup>25</sup> Asakura et al.<sup>25</sup> created an ROC (receiver operating characteristic) curve and reported that the cut-off value of the "self-report" version of the LSAS-J was 44 points. In the present study, a total LSAS-J score of 44 points or more was judged as indicative of an individual with SAD.

### *Overview of SLT*

Before the training began, the physicians carefully identified the cases' physical and psychological symptoms. Cases were also questioned about the nature of their stuttering. In all the cases, training was delivered by speech-language-hearing therapists with more than 5 years of clinical experience treating stuttering. The aim of the training was to learn fluency skills in the training room using fluency-shaping therapy, and then use these

skills outside the training room in everyday speaking situations. To be clear, the aim of the training cannot be achieved if the patients do not attempt their learned fluency skills in everyday speaking situations. Patients were encouraged to try to use their fluency skills outside the training room in graded steps starting from a simple situation, and, as much as possible, not to avoid speaking situations, not to substitute words, and not to rephrase what they wanted to say. The patients were also required to take control of the practice themselves, and use their newly learned fluency skills in everyday life to support their retention. The training sessions lasted from 40–60 minutes and were conducted once or twice a month.

The training content was as follows. First, the patient was shown how speech works (the various mechanics of breathing, vocalization, and articulation) and the characteristics of speech production by those who stutter. Next, fluency skills that allow stuttering to be reduced were described using training materials. There are three fluency skills: lengthening the duration of the indrawn breath before vocal attack, soft voice (whereby acoustic pressure at the time of vocal attack is reduced), and reduction of the discontinuity of the voice by lengthening the duration of the vowel sound during vocalization. Cases with severe symptoms, who found it hard to practice fluency skills, first practiced modulating the speed of their speech when reading a passage aloud at a slow speed of 2 syllables per second, before progressing to the fluency skills.

The training exercises comprised basic exercises and applied exercises. In the basic exercises, the speech-language-hearing therapist instilled fluency skills into the patient in the training room through imitation using: 1) single syllables (50 sounds), 2) expressions (words including proper nouns, such as names and addresses, formulaic greeting phrases, numbers, and words that the patient found hard), 3) reading aloud (50–100 clauses), 4) monologs, and 5) various conversational situations. In the applied exercises that followed, everyday speaking situations that the patient found hard were imagined. The therapist and the patient practiced using fluency skills in one-to-one role-plays in such situations, which could include: 1) a presentation in an academic or work setting, 2) making or receiving a telephone call, and 3) a face-to-face job interview or one for a position in an academic institution.

The patients' questions about stuttering were answered with scientifically sound information in a way that was easy for them to understand. The therapists also had the opportunity to talk with the patients about their questions and concerns related to stuttering in everyday

communication, such as whether or not to tell those around you about your stuttering, when the therapist could then tell them various relevant opinions on stuttering. Moreover, the therapists also listened empathetically whenever the patients had anxiety crises about stuttering. If a therapist was worried that the patient's anxiety was increasing, they could report it in a timely manner to the physician in charge.

#### *Outcome of SLT*

Patients' SLT outcomes were classified into 3 groups: "completed," "ongoing," and "discontinued." The termination criteria for SLT in the present study were: (1) stuttering frequency was reduced or eliminated, (2) avoidance behavior was reduced or eliminated in daily speech situations, and (3) practice was self-managed. SLT was terminated when all 3 of these criteria were achieved. The methods for confirming whether each termination criterion was achieved were as follows.

First, it was judged that the stuttering frequency was reduced or disappeared when the stuttering frequency was less than 5% during the text reading period. Next, as a method for confirming the reduction or disappearance of avoidance, we evaluated how many days a week the patients consciously replaced difficult words with other words or consciously avoided difficult situations during each training session. Based on the patient's self-report, a speech-language-hearing therapist evaluated this on a 5-point scale (score: 0 [part of 1 day], 1 [1–2 days], 2 [3–4 days], 3 [5–6 days], 4 [every day]). When the score was 0 or 1, it was determined that the avoidance behavior had reduced or disappeared.

Finally, as a method of confirming the patient's self-management of practicing these fluency skills, the number of practice days in the immediately preceding week was evaluated for each training period. Based on the patient's

self-report, a speech-language-hearing therapist evaluated the report on a 5-point scale (score: 0 [every day], 1 [5–6 days], 2 [3–4 days], 3 [1–2 days], 4 [part of 1 day]). If the score was 0 or 1, it was judged that the exercise was self-managed.

#### *Statistical analyses*

The total LSAS-J score and average stuttering frequency were compared using a *t*-test. The correlation between stuttering frequency and total LSAS-J score; age and total LSAS-J score; and age and stuttering frequency were examined using the Pearson's correlation coefficient. The percentage of clinical outcomes of SLT and the rate of achievement of the termination criteria were tested for the difference in ratios using Fisher's exact test. The significance level was set to 5% ( $P < 0.05$ ), and SPSS, version 24, was used for the statistical analyses.

## **Results**

#### *Patients' characteristics*

Table 1 shows the patients' characteristics. The average of the total LSAS-J score of all 88 cases was  $47.5 \pm 25.5$  points, and the frequency of stuttering during the text reading period was  $8.6 \pm 7.7\%$ . Six of the 88 cases (6.8%) had a high total LSAS-J score of 90 points or more. Further, the average total LSAS-J score was  $44.1 \pm 23.9$  for males and  $57.5 \pm 28.1$  for females, and females had significantly higher scores than did males ( $P < 0.01$ ). The average stuttering frequency was  $8.5 \pm 7.6\%$  for men and  $8.6 \pm 7.9\%$  for women, which was not significantly different ( $P = 0.97$ ). No correlation was found between stuttering frequency and the total LSAS-J score ( $r < 0.01$ ). Furthermore, there was no correlation between age and total LSAS-J score, or age, and stuttering frequency ( $r = 0.03$ ,  $r < 0.01$ , respectively).

**Table 1.** Patient characteristics

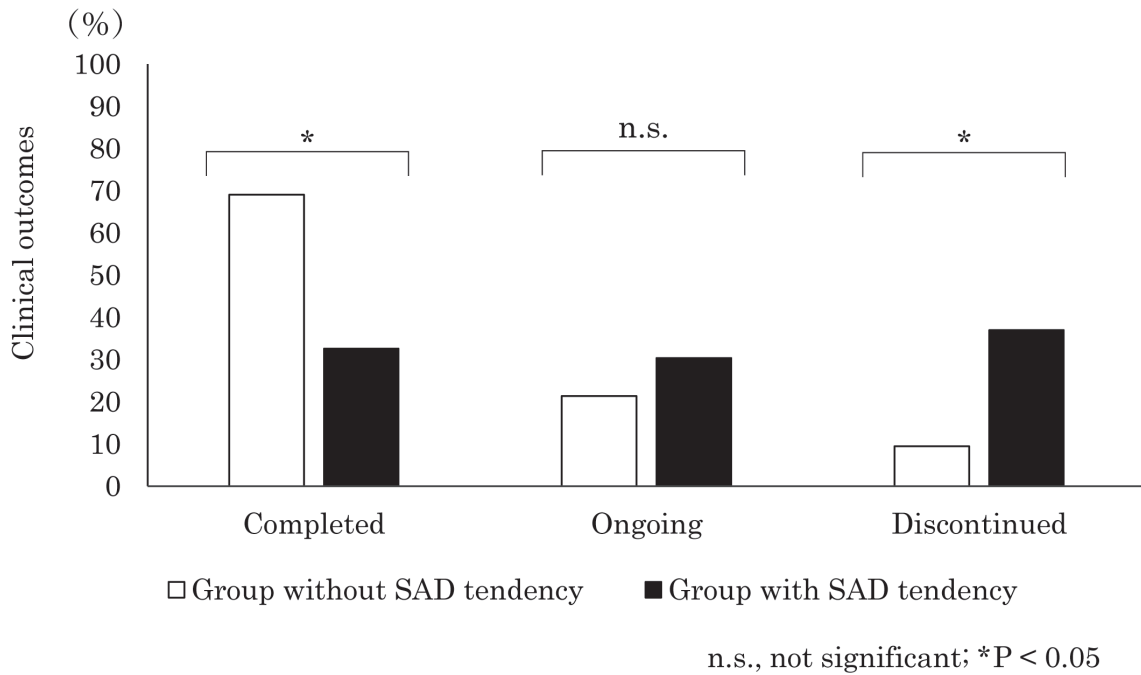
	All patients (n = 88)	Group without SAD tendency (n = 42)	Group with SAD tendency (n = 46)
Age mean (SD) years	26.7 (7.2)	26.2 (6.0)	27.2 (8.2)
Sex number (%) male	66 (75.0)	34 (81.0)	32 (69.6)
Stuttering frequency mean (SD) %	8.6 (7.7)	8.8 (7.9)	8.3 (7.5)
LSAS-J total score mean (SD) points	47.5 (25.5)	26.2 (11.1)	66.9 (18.4)

When all 88 cases were classified into 2 groups based on the LSAS-J cutoff value, 46 cases (52.3%) had SAD (the group with SAD), and 42 cases (47.7%) were without SAD (the group without SAD). The total LSAS-J score was  $66.9 \pm 18.4$  points for the group with SAD and  $26.2 \pm 11.1$  points for the group without SAD. The stuttering frequency was  $8.3 \pm 7.5\%$  in the group with SAD and

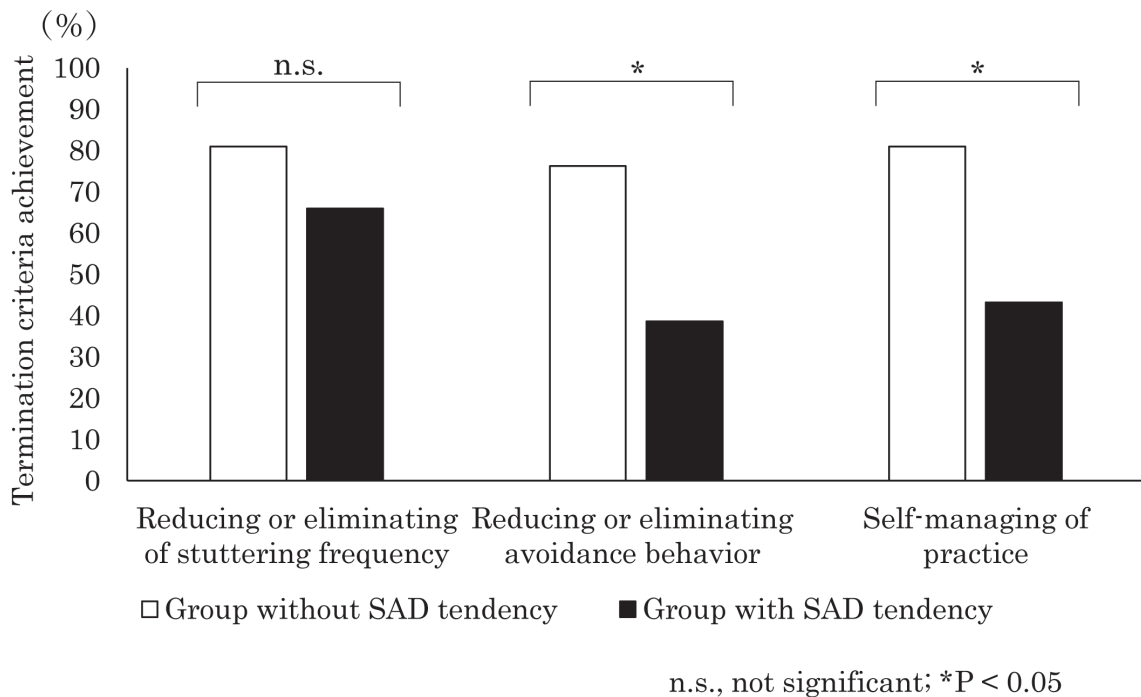
$8.8 \pm 7.9\%$  in the group without SAD. No significant difference was observed between the two groups ( $P = 0.75$ ).

*Outcomes of SLT with and without SAD*

Six months after the commencement of SLT, of the 88 cases, 44 patients (50.0%) completed training, 23 (26.1%)



**Figure 1.** The percentage of clinical outcomes at 6 months after the commencement of SLT with or without SAD



**Figure 2.** The achievement rate of termination criteria at 6 months after the commencement of the SLT with or without SAD

continued training, and 21 patients (23.9%) discontinued training.

Further, among the 42 patients without SAD, 29 (69.1%) completed training, 9 (21.4%) continued training, and 4 patients (9.5%) discontinued training. On the other hand, of the 46 patients with SAD, training was completed in 15 (32.6%), continued in 14 (30.4%), and discontinued in 17 patients (7.0%). The rate of completion of training was significantly higher in the group without SAD than that in the group with SAD [ $\chi^2(1) = 11.66, P < 0.01$ ]. The rate of discontinuation of training was significantly lower in the group without SAD than in the group with SAD [ $\chi^2(1) = 9.09, P < 0.01$ ]. There was no significant difference in the rate of continued training between the 2 groups [ $\chi^2(1) = 0.92, P = 0.37$ ] (Figure 1).

Regarding the achievement rate of the three termination criteria at 6 months after the commencement of SLT, among 42 patients without SAD tendency, 34 (81.0%) had reduced stuttering frequency, and 32 patients (76.2%) had reduced or disappearance of the avoidance behavior, moreover, 34 patients (81.0%) were able to self-manage the exercises. On the other hand, among 44 patients with SAD, stuttering frequency decreased in 29 patients (65.9%), avoidance behavior decreased or disappeared in 17 patients (38.6%), and the practice of self-administration was observed in 19 patients (43.2%). There was no significant difference in the rate of decreasing stuttering frequency between the 2 groups [ $\chi^2(1) = 2.48, P = 0.12$ ], but the rate of decrease or disappearance of the avoidance behavior [ $\chi^2(1) = 12.36, P < 0.01$ ], and the ratio of those who were able to self-manage the practice exercises [ $\chi^2(1) = 12.96, P < 0.01$ ] was significantly different (Figure 2).

#### *Changes in SAD between the groups with completed and ongoing training*

In the completed-training group (44 cases) and the ongoing-training group (23 cases), we compared the changes in the LSAS-J scores at the first visit of the training and 6 months after the commencement of SLT. The total LSAS-J score at the first visit of the ongoing-training group (average  $55.2 \pm 25.6$  points) was significantly higher than that of the completed-training group (average  $35.1 \pm 20.0$  points,  $P < 0.01$ ). The ratio of patients with the LSAS-J score at the cutoff value or higher at the first visit was 15 of 44 patients (44.1%) in the completed-training group and 14 of 23 patients (60.9%) in the ongoing-training group. The percentage of those who had SAD was significantly higher in the ongoing-training group than that in the completed-training group ( $P < 0.01$ ).

The total LSAS-J score in the completed-training group at the end of training was  $24.4 \pm 12.0$  on average, which was significantly lower than that at the first visit ( $P < 0.01$ ). Although 2 of 44 patients (4.5%) had SAD above the LSAS-J cutoff value at the end of the training, the ratio of patients with the LSAS-J score at the cutoff value or higher was significantly decreased compared to the first visit of the training [ $\chi^2(1) = 12.32, P < 0.01$ ].

On the other hand, the total LSAS-J score in the ongoing-training group at 6 months after the commencement of SLT was an average of  $49.2 \pm 24.4$ , and no significant difference was observed compared to that at the first visit ( $P = 0.42$ ). The number of patients who suffered SAD above the LSAS-J cutoff value was 11 patients (47.8%) at 6 months after the therapy, and there was no significant difference compared with that at the first visit [ $\chi^2(1) = 0.79, P = 0.37$ ].

## **Discussion**

### *Patients' characteristics*

Approximately 50% of adult stutterers in the present study who visited a medical institution had SAD by screening. This was similar to an overseas report that 40% of adult stutterers had SAD.<sup>13</sup> The reason for the slightly higher numerical value was considered to be the effect of the subject attributes. The subjects in this study were patients who suffered from stuttering and visited a medical institution. Of the total patients, 6.8% included those with a total LSAS-J score of 90 or more and severe SAD. This may have reflected the low consultation rate at medical institutions among the SAD patients.<sup>25</sup> No correlation was found between stuttering frequency and the total LSAS-J score. It has been reported that there is no correlation between the objective severity of stuttering and the presence or absence of SAD.<sup>15</sup> The results of our study support this previous study.

There was no correlation between age and the total LSAS-J score, and between age and stuttering frequency. Therefore, when dealing with people with stuttering who visit medical institutions, it is necessary to keep in mind that there may be persons with a high SAD tendency and severe stuttering symptoms regardless of age. As for the relationship with gender, females had a higher SAD frequency than did males, and this was also similar to the results of a previous study.<sup>26</sup> Therefore, it is considered that the sample in this study is similar to the previously reported relationship between stuttering and SAD.

### *Outcomes of SLT with and without SAD*

We performed SLT with the goal of using fluency shaping

skills to teach how to easily stutter less in a training room and to generalize that skill into everyday speech situations. About two-thirds of the group without SAD were able to finish the training within the first 6 months. Therefore, training using the fluency shaping approach proved suitable for stutterers without SAD. On the other hand, there were patients without SAD who continued training even after 6 months of the commencement of therapy. One of the factors was considered to be the effect of stuttering severity. No correlation has been reported between the degree of SAD and the objective severity of stuttering.<sup>14</sup> Therefore, it is possible that some people with severe stuttering were included in the group without SAD, and it was speculated that a longer training period was required due to severe stuttering.

Only one-third of the patients in the group with SAD completed the training 6 months after the commencement of training. Overseas, it has been reported that those with both stuttering and mental illness have longer training periods.<sup>27</sup> Therefore, it is considered that one of the reasons the number of patients who completed the training was small was that SAD had an effect on the duration of SLT using the fluency shaping approach. Fluency shaping skills can be easily used repeatedly in speech therapy situations (e.g., the training room), but it is difficult to generalize it to everyday speech situations (e.g., the outside the training room). Our results indicate the possibility that SAD caused anxiety, fear, and evasive behavior in the utterance scene, which made it difficult to continue the practice in the everyday utterance scenes and, therefore, prolonged the training period. In addition, about one-third of the group with SAD discontinued training. There are several possible reasons for their discontinuation. It goes without saying that the training method may not be suitable for each and every individual. Although it was a single case, the treatment report for a stutterer with SAD was successful in reducing stuttering symptoms and SAD symptoms using SLT and drug therapy.<sup>28</sup> Furthermore, it has been reported that group therapy using the stuttering modification method, which is one of the direct methods for stuttering, improved the psychological index.<sup>29</sup> Although various treatments are used alone or in combination as described above, there is currently no clear selection criterion or "gold standard," for the treatment method for stutterers with SAD. Therefore, when a speech-language-hearing therapist treats stutterers with SAD, it will be necessary to prepare multiple treatment options in advance and explore the method suitable for each stutterer.<sup>29,30</sup>

#### *Change of SAD in the completed-training and the ongoing-training groups*

In the completed-training group, social anxiety was alleviated at the end of the training as compared to that at the first visit. This could be attributed to a number of factors. First, it is possible that SLT reduced social anxiety. Menzies et al.<sup>31</sup> mentioned the use of exposure therapy by speech-language-hearing therapists to stimulate generalization of fluent speech to daily life within typical adult stuttering clinical practice. In this study, speech-language-hearing therapists encouraged the use of fluency skills in everyday life, and it was possible that this process amounted to an "exposure" experience and reduced social anxiety. Second, it is possible that through self-administration of the LSAS-J questionnaire, cases with stuttering improved their metacognition regarding the degree and nature of their own social anxiety. This implies that the act of objectively assigning a numerical value to the degree and nature of their own social anxiety motivated the patients to somewhat reduce avoidance behavior. A third possible explanation could be that, thanks to the sympathetic involvement of speech-language-hearing therapists in listening to the distress of patients with stuttering, the patients themselves came to terms with their stuttering. The importance of counseling patients with stuttering has been documented.<sup>32</sup>

However, for the ongoing-training group, no change was found in the LSAS-J overall score 6 months after the commencement of training, which could probably reflect the low level of SAD's natural remission rate.<sup>33</sup> Thus, it could be important to re-examine the appropriateness of LSAS-J scores for patients with stuttering and to confirm any changes in their social anxiety.

#### *Future prospects*

The patients in this study were adult stutterers 18 years of age and older. Approximately 75% of SAD cases develop during school age, at around 13 years old.<sup>1</sup> Therefore, it is necessary to evaluate the presence or absence of SAD in adolescents. However, in Japan, SAD in school-age stutterers has not been fully investigated, and further studies are warranted, including those assessing the effect of SAD on the duration of SLT for adolescent stutterers.

In addition, Autism spectrum disorder without mental retardation and attention deficit hyperactivity have been reported to coexist with SAD regardless of stuttering.<sup>34-35</sup> Stuttering and neurodevelopmental disorders coexisted in 3 patients in this study. Therefore, the relationship between SAD and SLT in stutterers with these neurodevelopmental disorders warrants further examination.

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**Conflicts of Interest:** None

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