

Psychiatric comorbidities in adult patients who stutter

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Objectives: We sought to explore the psychiatric comorbidities and associated factors in adult patients who stutter.

Methods: A total of 195 adults who stutter (AWS) aged 18–64 years were recruited on their first visit, diagnosed by a child psychiatrist, and grouped according to their comorbidities. Psychological measures: Patient's Health Questionnaire-9 (PHQ-9), Liebowitz Social Anxiety Scale Japanese version (LSAS-J), the Modified Erickson Scale of Communication Attitudes (Erickson S-24) were compared using the mean scores among groups. Demographic and clinical information was also compared.

Results: Of 195 AWS seeking treatment for stuttering, 52.3% had psychiatric comorbidities, 29.2% had depression and/or anxiety disorders, 26.6% had autism spectrum disorder (ASD), 13.3% had overlapping psychiatric diagnoses. Group comparison was done in 169 patients grouped into 3 comorbidity groups: stuttering only, depressive/neurotic (D/N), and ASD groups. The PHQ-9 and Erickson S-24 scores differed significantly ($P < 0.000$, $P < 0.024$, respectively). The mean score of the LSAS-J for AWS was high, and the D/N group had a significantly higher mean score compared to that of the SO group. Other factors such as stuttering severity and age did not differ significantly between groups, however, "highest education received" and "present social status" did differ significantly ($P = 0.000$, $P = 0.024$, respectively).

Conclusion: Clinicians should keep in mind that psychiatric disorders are highly comorbid in treatment-seeking AWS and that ASD is one of the most prevalent comorbidities among them.

Key words: stuttering, comorbidity, autism spectrum disorder, social anxiety, PHQ-9

Abbreviations: AWS, adult(s) who stutter; PHQ-9, Patient's Health Questionnaire-9; LSAS-J, Liebowitz Social Anxiety Scale Japanese version; Erickson S-24, Modified Erickson Scale of Communication Attitudes; ANOVA, analyses of variance; ASD, autism spectrum disorder; PWS; person (or people) who stutters; SAD, social anxiety disorder; NRCD, National Rehabilitation Center for Persons with Disabilities; ASC, Adult Stuttering Clinic; SD, standard deviation; D/N, depressive/neurotic; SO, stuttering only; OCD, obsessive-compulsive disorder; GAD, generalized anxiety disorder; PRIME-MD, Primary Care Evaluation of Mental Disorders; PARS-TR, Parent-Interview ASD Rating Scale-Text Revision

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Introduction

Stuttering is a disorder in verbal communication or fluency. Onset of stuttering is early during the toddler age, the peak onset being 2–3 years old, with well-known overt symptoms, such as syllable repetitions, prolongation, blocking, and stoppage of sounds. Though stuttering is estimated to be found in about 5%–8% of children, those who persist in stuttering until into adulthood are estimated to be around 1% of the adult population.¹ Fluency disorders including stuttering are partly thought to be developmentally natural phenomena accompanying the explosively growing vocabulary and speech ability exceeding the physical ability to produce speech in toddlers.¹ Natural remission of stuttering may occur with growth, but those whose stuttering did not remit would likely experience considerable emotional stress in their adolescence.² Because most patients become able to anticipate stuttering, attempts to cope with stuttering by pausing, omitting, substituting, circumlocuting or avoiding the word, or avoiding the speech situation itself, takes place and overt symptoms such as repetition of syllables become less dominant.³ This "progression of stuttering" enables stuttering symptoms to be less perceivable by others, but for the PWS, it often generates negative emotions, such as anxiety, reduced confidence, embarrassment, isolation, helplessness, denial of self, and frustration.^{2,4} It is reported that PWS continue stuttering through adolescence and often hide the fact even to their families.⁴

SAD

SAD which is reported to exist in $\leq 40\%$ of adults who stutter (AWS), whereas the prevalence in general population is estimated to be approximately 4%–15%.^{5–7} Psychiatric disorders have been reported in 38% of adolescents seeking speech therapy for stuttering.⁵ SAD is described in the Diagnostic and Statistical Manual of Mental Disorders 5th edition (DSM-5),⁸ is a disorder defined by marked or intense fear of social or performance-based situations where scrutiny or evaluation by others may occur. Though they know that their fear of negative evaluation is overestimated and extreme, it causes significant mental distress, decreasing the quality of life. In PWS, anticipated anxiety and fear for speech situations is an understandable reaction formed by actual experiences of having been teased or bullied for stuttering.⁴ SAD is diagnosed in PWS when the extent of anxiety in social situations is disproportionate even taking into account the past negative experiences

associated with the patient's extent of anxiety.⁹

Chronic stress due to disability or disease is a risk factor for psychiatric disorders such as anxiety and depression.¹⁰ Chronic anxiety is known to be a high risk factor for developing depression, and PWS comorbid with SAD or any other anxiety disorders may well be at the high end of the risk spectrum. Psychiatric disorders in PWS have been studied, but mostly in relation to SAD. Depressive disorders and other psychiatric disorders among PWS have been studied far less.

The primary aim of this study was to discover the prevalence of psychiatric comorbidities especially depressive disorders in AWS by directly interviewing patients. The secondary aim was to examine whether or not there were any factors associated with psychiatric comorbidities in AWS in Japan.

Subjects and Methods

The NRCD has an ASC for patients 18–64 years old for treatment of stuttering. Appointment is made on the phone. There is typically a 2–3 months waiting period until the first appointment. On the first visit, patients fill-in several questionnaires in the waiting room, which normally takes approximately 1–2 hours. In the examination room, the patient is examined by an otolaryngologist who interviews the patient and conducts the necessary physical examinations. A speech therapist is usually also present in the examination room. The patient then moves to another room, with the speech therapist, and undergoes speech assessment. Based on the results of the physical examinations and the speech assessment, the treatment plan is then explained to the patient by the otolaryngologist. For the present study, we asked each patient whether or not they would agree on having a child psychiatrist present at the first visit interview and being asked questions, explaining the purpose of this study and how the data would be used and analyzed by providing written and oral information. A child psychiatrist attended each patient's first visit examination after he or she gave written informed consent. The admission sheets and the psychological questionnaires were scored and checked by the otolaryngologist, speech therapist, and the child psychiatrist before calling the patient in for the examination. After the interview by the otolaryngologist was completed, the child psychiatrist added questions in a semistructured psychological interview to screen for depressive or anxiety disorders or any developmental disorders. Diagnosis of psychiatric comorbidity was made according to the DSM-5.⁸ All 195 patients asked to

participate in this study consented.

The study was approved by the human research ethics committee of the NRCD, which both the research and clinical divisions, that treat children and adults who stutter, belong to. Written informed consent was given by the participants in accordance to the committee guidelines. Data for this study were collected over a 5-year period (2013–2017). Three participants who were not diagnosed as AWS were excluded, leaving a total of 195 participants for the study.

Measures

Diagnosis of psychiatric comorbidities

Diagnosis was made by a child psychiatrist who regularly sees patients of a broad age range who have various comorbidities who were referred to us from other clinics, educational institutions, or occupational health physicians, for assessment of developmental disorders. Several questions were added by the child psychiatrist in a semi-structured way to screen for present or past psychiatric disorders at the first visit examination at the ASC. Both closed and open questions were asked, such as the following. "Do you think you worry more than others?" "Have you felt so low that you were absent from school or missed work?" "Have you ever felt so depressed as to think about committing suicide?" "When were you the happiest in the past?" "Have you ever talked about your stuttering with someone?" "Have you ever been told that your language development was slow?" "What kind of a child were you in elementary school?" "Were you often scolded for forgetting homework or something for school?" "How is your sleep?" "Is it easy for you to fall asleep?" "How do spend your free time?"

In many cases, the patient's parent(s) accompanied the patient to the first visit and provided additional information which was very helpful. Diagnosis was made according to the DSM-5.⁸ If the patient was currently not undergoing treatment, informing the patient in advance that the diagnosis was a tentative one, we recommended the patient visit a psychiatric clinic for treatment. If a developmental disorder had been newly diagnosed, we recommended receiving an assessment from, and informed the patient about our child psychiatry clinic, because psychiatric clinics that diagnose developmental disorders in adults are not yet common in Japan. The assessments at child psychiatric clinic included: the PARS-TR, the Wechsler test, and the Vineland Adaptive Behavior Scales, Second Edition.^{11,12} The PARS-TR is an interview-based rating scale widely used in Japan developed to screen for ASD, has been shown to have good discriminative validity in differentiating between ASD patients and nonclinical controls, regardless of intellectual capacity, and has good convergent validity with the ADI-R (Autism Diagnostic Interview-Revised).¹³ Even though a patient had already been diagnosed and was being treated at another clinic, similar questions were asked, and we made a diagnosis. If the patient already had a diagnosis of bipolar disorder or past history of schizophrenia, that diagnosis was adopted.

Depressive disorders and anxiety disorders including SAD or overlapping of the two disorders were all grouped together in a single category which we named, D/N. Whether anxiety disorders and depressive disorders should be understood as distinct disorders or a combined disorder has been, and is currently being discussed. An investigation of symptomatology from a large

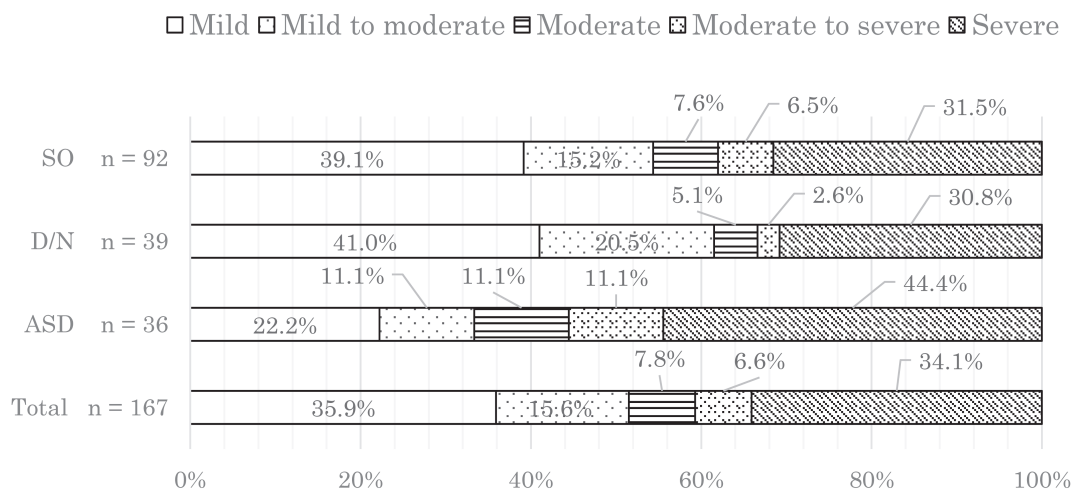


Figure 1. Distribution of stuttering severity in the psychiatric comorbidity groups of AWS

psychopathological network has shown direct and indirect associations between anxious and depressive symptoms.¹⁴ The high comorbidity rate of different anxiety disorders, and depressive and anxiety disorders have been reported.^{15,16} Childhood and adolescence research has shown that anxiety disorders and depressive disorders predict each other over time, and that the high comorbidity rate of both disorders could be partly explained by shared genetic risk factors.¹⁷ Primary care patients with depressive or anxiety symptoms are known to be of high risk of also having comorbid psychiatric disorders, and even a subthreshold of both symptoms has been reported to be equally or more detrimental to the patient's quality of life. Therefore, we considered that grouping both depressive and anxious patients together in the D/N group was acceptable from both a symptomatic and an etiologic point of view.

Patients' demographic data

Data were harvested from the interview admission sheets and questionnaires which asked about: present age and onset age of stuttering, job or social status, family members, family history of stuttering, history of visits to medical clinics, history of physical or mental disorders, history of treatment for stuttering, speech-class attendance at school, and present medications. However, complete sets of these data were not available from all of the participants.

Speech assessment

Two licensed speech therapists, who work at the clinic, did the assessments of stuttering severity as part of a routine, clinical, first visit procedure.

Objective assessment of stuttering

Overt stuttering symptoms, or objective stuttering symptoms were measured by the previous version of the Assessment of Stuttering¹⁸ which is currently used clinically throughout Japan. The Assessment of Stuttering scores included: the percentage of stuttered phrases, duration of stuttering, degree of bodily tensing while stuttering, degree of manifestation of accompanying symptoms at different speech situations, such as monologue, free conversation, reading word lists, reading aloud, and picture explanation. The test reveals stuttering severity in a profile style; however, in the present study, we adopted the highest percentage of observed stuttered phrases as the index of stuttering severity. According to the manual, the degree of stuttering corresponds to the measured percentage of stuttered phrases in the speech in the range of: mild, 3% – <5%; mild to moderate, 5%

– <10%; moderate, 10% – <15%; moderate to severe, 15% – <20%; severe, ≥20% (Figure 1).

Subjective assessment of stuttering

The self-rated subjective stuttering severity was rated in 5 categories: "not stuttering," "may not be stuttering," "mild," "moderate," and "severe," but there were no patients who scored "not stuttering." Self-evaluations of stuttering severity have been developed and used in treatment and research with PWS. There are more than 50 self-reporting protocols, of which Erickson S-24¹⁹ is one of the most well used, that evaluate various aspects of stuttering: anxiety, avoidance of speaking situations, and self-efficacy level.²⁰ Subjective stuttering severity is an essential measure for assessing stuttering severity and treatment outcome because covert symptoms such as avoidance or substitution of words, is not perceivable by objective measures.^{21,22}

Psychological data

The Erickson S-24 is a self-rating 24-item scale to evaluate communication attitudes.¹⁹ Patients grade self-perceived communication attitudes in various situations. The Erickson S-24 asks patients 11 questions to assess their communication attitudes of avoidance in various situations and 1 question to assess word avoidance. The remaining 12 questions ask patients about communication attitudes toward making a good impression, how they feel while talking, and their degree of self-control while talking. Higher scores indicate poorer communication attitudes and higher risk of relapse after speech therapy for stuttering.

The mean Erickson S-24 score for 200 patients with stuttering was reported to be 16.4 (standard deviation [SD] = 5.8) in a Japanese clinical sample.²³ It was reported that gender does not significantly effect the results of the Erickson S-24.^{23,24}

The LSAS is an interview form for measuring social anxiety symptoms which consists of 24 items each of which asks about the anxiety or fear the person would feel and the degree to which the person would avoid the situation on a 4-point Likert scale of 0 – 3 concerning specific socializing or performing situations. In a study of adult SAD patients measured by a clinician who interviewed the patients using the LSAS, a cut-off score of 30, and a cut-off score of 60 for a much severer subtype of SAD, categorized as GAD in the DSM-III-R or DSM-IV and DSM-5, were reported.²⁵ The LSAS-J is a Japanese version of the LSAS and has been confirmed to have a high reliability and validity.^{26,27} It had a high correlation with the Japanese version of the SADS (Social

Avoidance and Distress Scale) and high internal consistency and structural-construct validity, a cut-off point of 42 was confirmed, showing that the LSAS-J was a reliable measure of social anxiety symptoms in Japan.^{26,27} The mean score of the self-scored LSAS-J from a sample of 929 subjects who did not stutter as a control in Japan was reported to be 42.4 (SD = 27.5),²⁸ and a cut-off point of 44 has been suggested for the self-rated LSAS-J in the Japanese population.²⁷

For the present study, the PHQ-9 was newly added to the questionnaires routinely used for the Adult Stuttering Clinic to screen for depressive disorders. Patient Health Questionnaire (PHQ) is a self-administered version of the PRIME-MD which is a clinician-administered questionnaire developed by R. L. Spitzer, M.D., as a brief screening tool for psychiatric disorders commonly seen in primary care.^{29,30} The PHQ showed excellent diagnostic validity, similar to the original PRIME-MD.²⁹ The PHQ-9 is also a reliable measure to determine the severity of depression.³¹ The PHQ-9 consists of 9 questions from the PHQ and a cut-off point of 10 for primary care patients was recommended to discriminate moderate depression from non-depression.³¹ The diagnostic validity of the PHQ-9 was in agreement with the diagnoses suggested by mental health professionals using the Structured Clinical Interview for DSM-III-R, and has high correlation with the currently widely used clinical measures to determine the severity of depression such as the 17-items Hamilton Depression Rating Scale and the Montgomery-Åsberg Depression Rating Scale (MADRS, 10 items).³² The American Psychiatric

Association recommends using the PHQ-9 as an assessment tool for depressive disorders in its newly revised DSM-5.⁸ In a study with a large psychiatric population (n = 1,023), the PHQ-9 showed good sensitivity (0.83) and high specificity (0.72) at a cut-off point of 13, and was recommended for screening current major depressive symptoms in psychiatric outpatients.³³

Studies in Japan reported different cut-off scores of PHQ-9 (range, 5–14).^{34,35} In a study examining the utility of PHQ-9 in a Japanese primary care cohort, the PHQ-9 results were acquired from 521 patients, 42 patients were diagnosed as having major depressive disorder, a cut-off point of 11 was suggested (0.76 and 0.81, respectively).³⁵ There was no relation between PHQ-9 scores and age.³⁵

Statistical analyses

Demographic variables and psychological measures and stuttering parameters were compared between the 3 comorbidity groups. For continuous variables, comparisons were based on one-way ANOVA between groups. For the LSAS-J scores, Dunnett's *t* test was carried out as a post hoc test with the SO group as the reference group. The Erickson S-24 and PHQ-9 scores did not follow a normal distribution, therefore non-parametric analysis with the Kruskal-Wallis test was carried out for comparisons of mean scores. For ordinal scales such as stuttering severity the Kruskal-Wallis test was carried out. For categorical variables, χ^2 tests of independence were carried out for comparisons between groups. We used SPSS v23 for data analysis, and P values <0.05

Table 1. Psychiatric comorbidities diagnosed in AWS (N = 195)

	NPC	Dep	Anx	ASD	BPD	ID	Sz	OCD	ADHD	Total
NPC	93	0	0	0	0	0	0	0	0	93
Dep	0	13 ^a	7	8	0	1	0	0	0	29
Anx	0	7	20 ^a	5 ^b	1	2 ^b	0	2 ^b	1	38 ^b
ASD	0	8	5 ^b	36 ^a	1	3 ^b	2	1	0	56 ^b
BPD	0	0	1	1	3 ^a	0	0	0	0	5
ID	0	1	2 ^b	3 ^b	0	0 ^a	1	0	0	7 ^b
Sz	0	0	0	2	0	1	0 ^a	0	0	3 ^b
OCD	0	0	2 ^b	1	0	0	0	4 ^a	0	7 ^b
ADHD	0	0	1	0	0	0	0	0	0 ^a	1
Total	93	29	38 ^b	56 ^b	5	7 ^b	3 ^b	7 ^b	1	239 ^b

NPC, no psychiatric comorbidities; Dep, depression; Anx, anxiety disorder; BPD, bipolar disorder; ID, intellectual disability; Sz, schizophrenia; ADHD, attention deficit hyperactivity disorder

^aThe number of AWS with only 1 psychiatric diagnosis

^bThe sum of the patient numbers for the patients with psychiatric comorbidities is greater than the total number of patients because some patients have multiple comorbidities.

were considered statistically significant.

Results

Psychiatric diagnoses in AWS

Of the 195 AWS, 64 patients (32.8%) had a history of psychiatric disorder or were currently being treated at psychiatric clinics. Table 1 shows the number of patients with the new psychiatric diagnosis among the 195 AWS. Anxiety disorders were diagnosed in 35 (17.9%) patients. Depressive disorders were diagnosed in 29 (14.9%) patients, and there were 7 (3.58%) patients diagnosed with both depressive and anxiety disorders. AWS diagnosed with comorbid ASD totaled 52 (26.7%).

ASD

Among those 52 (26.7%) ASD patients, 16 (30.8%) patients were also diagnosed with other psychiatric disorders, such as depressive disorders (n = 8), anxiety disorders (n = 5), bipolar disorder (n = 1), schizophrenia (n = 2), and intellectual disorder (n = 3) (Table 1). Of the 52 AWS with comorbid ASD, 21 (40.4%) patients had a history of psychiatric treatments; however, only 4 (19.0%) of them had been diagnosed as having ASD, and 14 (66.7%) patients had been diagnosed as having either anxiety or depressive disorders. Later, during this study

period, 21 (40.4%) patients newly diagnosed as having ASD were examined at our child psychiatric clinic, where their ASD diagnosis was confirmed.

As many as 93 patients (47.7%) had no other psychiatric comorbidities and were assigned to the SO group. We excluded patients with more than 2 psychiatric diagnoses, other depressive and anxiety disorders, from the comorbidity groups; patients with a diagnosis of depression and ASD, or anxiety and ASD, were excluded from both the D/N group and the ASD group. Patients who had either OCD, intellectual disability, schizophrenia, or bipolar disorder were also excluded, because the number of cases was too small to make a new group; however, including them into either the D/N or ASD groups lacked epidemiological grounds. As a result, from 195 AWS, we formed 3 comorbidity groups: the SO group (n = 93), the D/N group (n = 40), and the ASD group (n = 36). Thus, 169 patients were assigned to one of the groups, and the remaining 26 patients were excluded from these analyses.

Demographic factors and variables

Demographic factors for each comorbidity group are shown in Table 2. The patients' ages ranged from 18–64 years, mean age 29.2 (SD = 9.2) years. Male-to-female ratio was 5.1:1, slightly higher than the reported

Table 2. Demographic data for psychiatric comorbidity groups in AWS

Demographic factors	SO (n = 93)	D/N (n = 40)	ASD (n = 36)	Total (n = 169)	P value
Sex					0.013 ^a
Male, n (%)	81 (87.1)	21 (67.5)	32 (88.9)	140 (82.8)	
Female, n (%)	12 (12.9)	13 (32.5)	4 (11.1)	29 (17.2)	
Age (years)					0.097 ^b
Mean (SD)	29.7 (9.9)	30.1 (7.4)	26.1 (8.4)	29.0 (9.4)	
Median	26.0	29.5	22.5	26.0	
Employment					0.000 ^{**c}
Employed, n (%)	57 (61.3)	23 (57.5)	10 (27.8)	90 (53.3)	
Student, n (%)	27 (29.0)	4 (10.0)	17 (47.2)	48 (28.4)	
Unemployed, n (%)	7 (7.5)	11 (27.5)	9 (25.0)	31 (18.3)	
Education					0.009 ^{**d}
High school, n (%)	19 (22.9)	20 (51.3)	14 (38.9)	53 (33.5)	
College or higher, n (%)	64 (77.1)	19 (48.7)	22 (61.1)	105 (66.5)	

*P < 0.05

**P < 0.01

^a χ^2 (2) = 8.734

^bF (2) = 2.370 Dunnett's test (P = 0.044)

^c χ^2 (2) = 23.970

^d χ^2 (2) = 9.525

epidemiological data of 4:1.¹

A significant difference was found in gender, employment status, educational background [$\chi^2(2) = 8.734, P < 0.05, \chi^2(4) = 23.970, P < 0.05, \chi^2(2) = 9.525, P < 0.01$, respectively]. The male to female ratio was higher in the ASD group and lower in the D/N group. The mean age was lowest, and the ratio of student status was highest in the ASD group, reflecting the high proportion of college students in that group (Table 2).

Speech variables

There were no significant differences in the patients' mean age at onset of stuttering among the 3 comorbidity groups: 7.56 (SD = 5.40), 8.49 (SD = 6.06), and 7.29 (SD = 5.55) years for the SO, D/N, ASD groups, respectively.

History of enrollment in speech classes was available from 154 patients. Forty-one patients had enrolled in speech classes during elementary school. The rate of those who had attended speech classes did not differ

among the 3 comorbidity groups.

The distributions of stuttering severity and subjective stuttering severity in the 3 comorbidity groups are shown in Figures 1 and 2, respectively, with no significant differences.

Psychological variables

Table 3 shows the mean scores, SD of self-rated checklists or scales for speech related anxiety (by the Erickson S-24), social anxiety symptoms (by the LSAS-J), and depressive symptoms (by the PHQ-9) for the 3 comorbidity groups.

The mean scores of the PHQ-9 for the 3 comorbidity groups were all below the cut-off score of 11; however, the mean scores differed significantly between the groups ($P = 0.000$). Moreover, the D/N and ASD groups had higher mean scores than that of the SO group (Table 3).

The Erickson S-24 mean score was 18.6 (SD = 4.0) (Table 3), which was similarly high compared to the

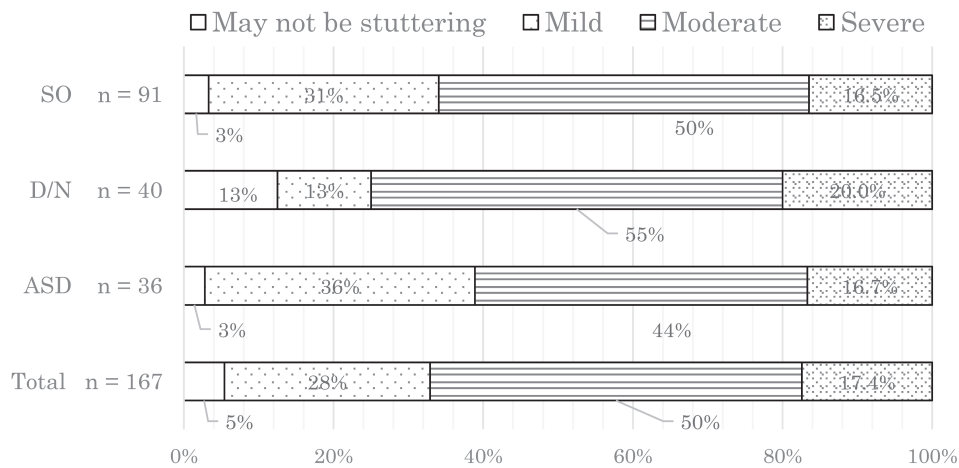


Figure 2. Distribution of subjective stuttering severity in the 3 psychiatric comorbidity groups of AWS

Table 3. Mean scores of psychological measures for the psychiatric comorbidity groups of AWS

Questionnaire (range)	SO (n = 92)	D/N (n = 40)	ASD (n = 35)	Total (n = 167)	P value
Erickson S-24 (0–24)					
Mean (SD)	17.9 (3.8)	19.1 (4.7)	18.9 (3.6)	18.4 (4.0)	0.024 ^a
LSAS-J (0–144)					
Mean (SD)	47.5 (23.9)	58.1 (30.5)	54.8 (26.4)	51.5 (26.4)	0.034 ^{a,b}
PHQ-9 (0–27)					
Mean (SD)	4.0 (4.4)	8.9 (6.3)	7.5 (6.0)	5.9 (5.7)	0.000 ^a

^aThe Kruskal-Wallis Test

^bDunnnett's *t* test

reported data from another sample of patients who stutter.⁴ The distribution of scores differed significantly between groups ($P = 0.024$). The D/N and ASD groups had higher mean scores compared to that of the SO group.

We used the Dunnett's test as a post hoc test comparing comorbid groups with the SO group control. The D/N group had a significantly higher mean score than did the SO group ($P = 0.034$) (Table 3). The mean score 47.5 (SD = 23.8) of the SO group was above the cut-off point of 44 for the self-scored LSAS-J in a Japanese psychiatric population (Table 3).²⁷

Discussion

The clinical sample of AWS in this study had a slightly higher male to female ratio than that in the reported epidemiological data of 4:1.¹ Patients with ASD generally have a higher male to female ratio of 4:1 – 3:1.³⁶ A high prevalence of developmental disorders in PWS has also been reported.^{37,38} Smith et al. reported in a prospective cohort study of 4-year-old children followed-up to age 11, that the group of children whose stuttering persisted had higher mean anxiety scores and higher autism prevalence (20.0%) than did naturally remitted (4.3%) or non-stuttering (5.9%) cohort groups.³⁹ ASD was diagnosed in 52 (26.7%) of 195 AWS in the present study, which was similar to the prevalence in children reported by Smith et al.³⁹ Of the 52 patients diagnosed as having ASD in the present study, as many as 31 (59.6%) patients would not have been diagnosed as having ASD if it was not for their stuttering symptoms and their participation in the study. Patients with undiagnosed ASD have had trouble or difficulty at the workplace, in job interviews, and getting credit for discussions at colleges, among other factors, that made them decide to seek treatment for stuttering.

Overlapping of psychiatric diagnoses were seen in high proportions among patients with ASD. It has been well documented that patients with ASD have multiple and high comorbidity rates of disorders including psychiatric disorders.⁴⁰ In a study conducted by self-report measures and structured interviews for psychiatric diagnoses, of patients with ASD 79% met the criteria for 1 or more lifetime diagnoses for a psychiatric disorder compared with 48.8% without ASD. Depressive disorders (57.2%) and anxiety disorders (53.6%) including OCD (21.7%) and agoraphobia (21.0%) were the most common.⁴⁰ Female gender was a significant predictor of depressive disorders, lower age and more severe ASD symptoms were associated with comorbidity of anxiety disorders.⁴⁰ In a study of 100 youths (aged 7

– 16), who visited an outpatient clinic for treatment of anxiety disorders, 62% had ASD symptoms.⁴¹ These results suggest that ASD and anxiety disorders are both highly predictive of each other.

A study using the LSAS-J with college students ($N = 104$, mean age = 19.96 [SD = 0.91], and male:female = 1:4.2) in Japan reported a mean score of 61.5 (SD = 23.0) in which the male only mean score was 71.3 (SD = 29.8).⁴² Using the LSAS-J, Kikuchi et al.⁴³ studied 100 PWS (mean age = 24 [range, 10 – 57] years, male:female = 3.7:1) in Japan and reported that "younger age" and "female" were associated with higher scores. Females 10 – 19 years old scored the highest mean score of 92.6 (SD = 12.0), whereas the mean was 37.7 (SD = 6.3) for patients older than 30 years old.⁴³ The rate of those whose scores were higher than 50, which correspond to moderate or very severe SAD, was 50.8% in that sample.⁴³ The sample in the present study had a higher mean age and a slightly higher male ratio compared to those in the Kikuchi, et al. sample.⁴³ However, the percentage of those who scored higher than 50 in the LSAS-J was 51.6% therefore equally high. In a study of 62 high school students seeking treatment for stuttering, the mean LSAS-J score was reported to be 49.8 (SD = 26.3), in which 51.6% had scores higher than the cut-off score of 44, and 19.4% had diagnoses (including suspicious ones) of comorbid psychiatric disorders (including neurodevelopmental disorders) made by pediatricians or child psychiatrists, who revealed that the LSAS-J scores were higher in comorbid cases.⁴⁴ The sample in the present study of 195 AWS had a high rate of psychiatric comorbidities. As many as 102 (52.3%) were diagnosed (including tentative diagnoses) with comorbid psychiatric disorders. The high comorbidity rate may be due to the bias of sampling or the high sensitivity for detection of ASD in this study.

Among AWS, the scores of PHQ-9 were higher in the comorbidity groups (the D/N and ASD groups) even though the mean scores were all below the cut-off score for moderate depression. Because the cut-off score varies with the sample condition in Japan, the mean PHQ-9 score of the D/N group and that of the ASD group, being below the cut-off score for depression, could be interpreted in such a way as to suggest that AWS may tend to understand their negative emotions as an inevitable effect of their stuttering. That, in itself, is the assumption that led to formulating the criteria to exclude disorders with apparent symptoms such as stuttering and Parkinson's disease from the diagnosis of SAD in the previous editions of the DSM-5.^{9,45} Diagnostic overshadowing on the side of patients and clinicians may

have hindered earlier detection of comorbid SAD or ASD and depression. It is important for clinicians who treat AWS to pay careful attention to the high comorbidity of psychiatric disorders, especially ASD, because the comorbidity may likely be treatment resistant. For such patients, appropriate approaches for ASD recommended in the current guidelines would most likely become necessary.

Limitations

There are two limitations to the present study. First, the patients were a clinical sample of AWS. Those with more severe stuttering symptoms and more milder psychiatric symptoms would likely visit the ASC for treatment for stuttering. Severely depressed patients or those with SAD generally would not likely seek treatment. Even if AWS are aware of their psychiatric symptoms, they may not seek help unless it becomes troublesome, worrying, or some external force encouraged them to get a check-up or took them to a hospital or clinic. Some patients have admitted to staying home for a considerable length of time up to many years before visiting an ASC. Second, even if the AWS had SAD and visited an ASC, the patient might not talk about the symptoms. Perhaps the patient had gone to a hospital or clinic with the intention of receiving treatment for stuttering and not for SAD, leading to an insufficient diagnosis which likely missed SAD entirely.

Conclusions

We found a high prevalence of ASD among AWS in this study. The PHQ-9 may be useful for screening for comorbid psychiatric disorders, not only SAD but also others, including ASD, in AWS. In treating AWS, clinicians should pay extra careful attention to the possibility of the presence of any comorbid psychiatric disorders, especially that of ASD. Though a patient's stuttering may not be completely cured, treatment of patients with psychiatric disorders may improve the quality of life for many patients who stutter.⁴⁶ Undiagnosed ASD could be a high risk for treatment resistant psychiatric disorders, in which case the patient will likely be treated with better results after having received a formal diagnosis of ASD. It is highly important in the treatment of AWS to be aware of diagnostic overshadowing of patients who may have a diagnosis of SAD and/or ASD.

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Conflicts of Interest: None

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