ADDUCTOR SPASMODIC DYSPHONIA AND BOTULINUM TOXIN TREATMENT: THE EFFECT ON WELL-BEING

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Adductor spasmodic dysphonia (AdSD) is a controversial and enigmatic voice disorder. It is generally accepted that it has a neurologic, although undetermined, cause, and it is accompanied by much psychological and physical distress. In this prospective study, standardized psychometric tests were used to assess the personality characteristics and psychological and somatic well-being of 46 patients with AdSD. Moreover, the effect of botulinum toxin (Botox) treatment on their well-being was evaluated. No significant differences could be detected between patients and a representative norm group concerning 7 personality characteristics. Nevertheless, before treatment, there were significantly more psychological and somatic complaints. After establishment of a normal to near-normal voice with Botox injections, these complaints were reduced to normal levels — a finding suggesting these phenomena to be secondary to the voice disorder. These findings, and the normal personality characteristics, do not support a psychogenic cause of AdSD.

KEY WORDS — adductor spasmodic dysphonia, botulinum toxin, psychometry.

INTRODUCTION

Adductor spasmodic dysphonia (AdSD) is an uncommon and poorly understood voice disorder that was first described by Traube¹ in 1871. The vocal symptoms are characterized by difficulty with voice initiation, a strain-strangled, effortful phonation with voice breaks, and glottal fry. Remarkably, these symptoms are reduced or absent during whispering, speaking or singing in a falsetto register, and nonspeech vocalizations (laughing, yawning). Spasmodic dysphonia is initially intermittent and task- or situationspecific. The marked intermittency and functional specificity of the symptoms have suggested a psychological basis for this unusual disorder.

Initially, (adductor) spasmodic dysphonia was considered predominantly as a conversion symptom of a hysterical illness affecting the coordination of speaking and breathing.²⁻⁴ Heaver⁵ argued that spasmodic dysphonia especially affected persons with a hysterical and narcissistic personality structure. Berendes⁶ analyzed several psychological factors in 23 patients with spasmodic dysphonia. He concluded that this voice disorder had to be considered as a neurosis without hysterical characteristics. However, these studies could offer no objective support for the hypothesized contribution of psychological factors in the ontogeny of spasmodic dysphonia, because they were based on case histories or patient descriptions.

To bypass the questionable reliability of interviewers and observers, psychometric tests were applied to assess the psychological phenomena related to this enigmatic voice disorder.⁷⁻¹⁰ These studies showed elevated levels of depression and anxiety and negative attitudes toward communication in AdSD patients as compared to matched normal controls. In one study, elevated levels of somatic complaints were also found.⁸ Treatment with botulinum toxin (Botox) injections generally reduced the levels of depression and anxiety, and significant attitudinal improvement was observed. Whether these differences could be explained as a reaction to a chronic disorder or as being related to a possible psychogenic origin of spasmodic dysphonia remained unsolved.

Nowadays, spasmodic dysphonia is thought to be a physical disorder, an idea already proposed by Schnitzler¹¹ in 1874. This is supported by the association of spasmodic dysphonia with other focal and generalized dystonias or involuntary movements, such as myoclonus and tremor. It is now assumed that AdSD has a neurologic (undetermined) cause, and it is characterized as a disorder of central motor

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TABLE 1. DEMOGRAPHIC DATA					
	No.	Age (y)		Duration of Symptoms (y)	
		Mean	Range	Mean	Range
Women	32	47.9	19-80	8.9	1-29
Men	14	53.7	31-68	_7.9	1-31

processing resulting in focal laryngeal dystonia. As well as the abnormal voice, a variety of other abnormalities have been found on objective tests, including electroencephalography, evoked potentials, blink reflexes, vocal reaction times, and responses to vagal stimuli.¹²

In summary, AdSD is generally accepted as a physical voice disorder associated with several psychological and somatic complaints. Some studies on the psychological impact of AdSD have been performed. The results of these studies supported the need for further research into the role of emotional aspects of spasmodic dysphonia prior to intervention, their relationship to objective and subjective voice changes, and how therapeutic management affects the longterm outcome of patients with spasmodic dysphonia.¹⁰ At present, the role of general personality variables in relation to AdSD is unknown.

The aim of this prospective study was twofold: 1) to investigate the hypothesis that personality variables might underlie spasmodic dysphonia and 2) to determine whether symptoms of negative affectivity (depression and anxiety) and somatic complaints are consequences of the disease.

SUBJECTS AND METHODS

Forty-six consecutive patients (32 female and 14 male) with AdSD were included in this study. The demographic data are summarized in Table 1. On the basis of their medical history, none of the patients appeared to have psychiatric or neurologic disorders, particularly, no other dystonias or familial tremors. All patients had had symptoms of AdSD for more than 1 year and had no previous treatment with Botox.

The personality characteristics of the patients were assessed with the Dutch Personality Questionnaire (DPQ).¹³ This questionnaire was developed in the early 1970s in the Netherlands and was adapted from the California Psychological Inventory.¹⁴ Extensive validational data are available on this inventory. The reliability and validity of the DPQ have been proven to be satisfactory.^{13,15-19} The 132 items of the DPQ, answered on a 3-point scale (True, ?, and False), are non-overlappingly keyed in the following scales: 1) neuroticism (21 items); 2) social anxiety (15 items); 3) rigidity (25 items); 4) hostility (19 items); 5) egotism (16 items); 6) dominance (17 items); and 7) selfesteem (19 items). The 7 scales are not independent: the highest scale intercorrelations are approximately .45. Usually, 3 second-order factors are found: emotional stability versus neuroticism (high loadings of scale 7 and with opposite sign scale 1); introversion versus extraversion (high loadings of scale 2 and with opposite sign scale 6); and dogmatism versus friendliness (high loadings of scales 3, 4, and 5).²⁰ The raw scores on the 7 DPQ scales were used in this study.

The Hopkins Symptom Checklist²¹ (HSCL) was applied to assess and monitor the psychological and somatic complaints existing before and during Botox treatment. This checklist is a self-report symptom rating scale that a subject answers on a 4-point scale of distress ("not at all" to "extreme"). In answering the HSCL, an explicit temporal referent is provided in terms of "How have you felt during the past seven days including today?" This makes the HSCL suitable for measuring changes, eg, measuring the effect of therapy. The Dutch version of the HSCL consists of 57 symptoms and complaints and has the following scales: somatic well-being (HSCL-Somat, 8 items), psychological well-being (HSCL-Psych, 17 items), and total score (HSCL-Total, 57 items). In this study, the norm group of the general population was used as a reference. There is ample support for the reliability and validity of the HSCL.21

To substantiate the effect of Botox treatment on the changes in psychological and somatic complaints, we also determined the changes in vocal function through subjective (patient) and objective (observers) ratings. The subjective rating consisted of statements concerning the patient's experience of his or her voice quality and performance (intelligibility, effort, and fluency of speech). Perceptual evaluation of spontaneous speech was used to rate voice quality objectively. All voice samples were judged by 3 experienced observers who were familiar with both spasmodic dysphonia and a perceptual evaluation rating system derived from the GRBAS system.²² For the purposes of this study, we only used the "G" parameter (grade) to quantify the overall impression of severity of the dysphonia. Assessment of the degree of dysphonia was achieved by calculating the means of the scores of the 3 observers. Both subjective and objective ratings were performed on visual analog scales. On the score form, each parameter was scored on a continuous horizontal line of 10 cm from bad or extremely pathological on the left side to good or normal on the right side. The distance in centimeters from the left side measured the score.

This study was part of a larger prospective clinical trial in which different procedures of Botox injections were compared in AdSD patients.²³ All 46 patients

TABLE 2. SCORES ON DUTCH PERSONALITY OUESTIONNAIRE¹³

	Spasmodic Dysphonia Group (n = 46)	Dutch Norm Group (n = 5,686)
Neuroticism	13.2 ± 9.4	12.0 ± 7.9
Social anxiety	10.2 ± 8.7	10.9 ± 6.9
Rigidity	26.9 ± 9.9	28.4 ± 7.8
Hostility	16.5 ± 7.8	18.2 ± 6.7
Egotism	11.2 ± 5.0	12.6 ± 5.0
Dominance	14.6 ± 6.7	13.8 ± 5.8
Self-esteem	27.8 ± 6.3	28.0 ± 5.6
Data are mean t -test, $df = 45$.	\pm SD. None of scales different	ed significantly; 1-sample

underwent unilateral and bilateral injections. During the first procedure, 5 units (1.25 U/0.1 mL) of Botox were injected in the left thyroarytenoid muscle only. The second treatment was performed after the voice quality had returned to the preinjection level (established by the patient and by audio and video registrations). During this procedure, both thyroarytenoid muscles were injected, each with 2.5 units (1.25 U/ 0.1 mL) of Botox. Under electromyographic guidance, a monopolar 27-gauge 30-mm Teflon-coated needle was inserted percutaneously, through the cricothyroid membrane and into the left thyroarytenoid muscle during the first procedure, and into both thyroarytenoid muscles during the second procedure.

The choice for the next treatment depended on the patient's preference. The dose was adjusted by titrating efficacy and side effects. The end of the study was the moment at which the patient experienced his or her former "normal" voice. However, a normal voice could not always be established. In those cases, the end point was the optimal voice that could be achieved.

The questionnaires were administered at the following times. The moment the patients entered the study (pretreatment), they were asked to complete the DPQ, the HSCL, and the self-rating questionnaire for (subjective) judgment of the voice. At that time, recordings were made for perceptual (objective) evaluation of the voice quality. During the follow-up period, ie, 1 month after the unilateral injection, 1 month

Changes in patients' and observers' voice ratings (visual analog scale [VAS] scores) and Hopkins Symptom Checklist (HSCL)–Total²¹ scores over course of time. Means are given, with associated 95% confidence intervals. Changes over time were statistically significant (p < .001) according to mixed-model analysis of variance. 1 — moment before Botox injection; 2 — 1 month after unilateral Botox injections; 4 — moment of "best voice ever since."



	Spasmodic Dysphonia Group (n = 46)	Dutch Norm Group (n = 406)	<i>p</i> *
HSCL-Psych	12.6 ± 8.6	7.9 ± 6.2	.0005
HSCL-Somat	3.5 ± 2.6	2.1 ± 2.3	.0005
HSCL-Total	36.8 ± 20.0	22.3 ± 16.5	<.0001
HSCL — Hop well-being, So	okins Symptom Che mat — somatic wel	ecklist, Psych — ps 1-being.	sychological
Data are mean	± SD.		
*One-sample t-	test, $df = 45$.		

after the bilateral injection, and at the moment of the optimal voice (posttreatment), all tests except for the DPQ were performed again.

RESULTS

Table 2¹³ provides descriptive statistics for the DPQ scores of the 46 AdSD patients prior to treatment with Botox. There were no significant differences between the patients and the representative norm group of the Dutch population in the raw scores of the 7 scales of the DPQ. On the other hand, the HSCL showed differences across all 3 scales, patients having significantly elevated mean scores compared to the representative norm group (Table 3²¹).

The Figure²¹ shows the changes in the patients' and observers' voice ratings and HSCL-Total scores over the course of time. Both the visual analog scale scores of vocal function and the HSCL scores showed continuous improvement during Botox treatment. These findings are reflected in Table 4,²¹ as well. Compared to the mean pretreatment scores of the HSCL, there was a significant decrease after treatment. In fact, the mean scores of all 3 scales reached values within the normal range. Comparing the scores of the AdSD patients after treatment with the representative norm group, we found no significant differences (Table 5²¹).

DISCUSSION

The first goal of this study was to investigate the hypothesis that personality variables might underlie



VAS

score

	QUALITI		
	Pretreatment	Posttreatment	
Patient rating of voice quality ⁺	3.4 ± 1.4	8.1 ± 1.5	<.001
Observer rating of voice quality [†]	3.7 ± 1.7	9.2 ± 0.9	<.001
HSCL-Psych	12.6 ± 8.6	6.1 ± 6.4	<.001
HSCL-Somat	3.5 ± 2.6	2.2 ± 2.6	.005
HSCL-Total	36.8 ± 20.0	18.6 ± 18.4	<.001
Data are mean ± S *Paired <i>t</i> -test.	D.		

TABLE 4. PRETREATMENT AND POSTTREATMENT SCORES ON HSCL²¹ AND RATINGS OF VOICE

[†]Zero represents "very bad or extremely pathological" voice, and 10 represents "good or normal" voice.

AdSD. Using the DPQ, we found no significant differences between the AdSD patients and the representative norm group of the Dutch population. On the basis of the DPQ results, no distinction could be made between normal and AdSD subjects. Thus, either there are no typical personality traits associated with AdSD or the DPQ could not detect those personality attributes. As the DPQ is accepted as an accurate, comprehensive tool, it would seem that the first hypothesis (AdSD is not associated with typical personality traits) is justifiable.

Our findings parallel the early work of Aronson et al,⁷ who applied the Minnesota Multiphasic Personality Inventory (MMPI) to 33 patients with spasmodic dysphonia. None of the 10 clinical scales of the MMPI could differentiate between the spasmodic dysphonia patients and a general medical outpatient population. On the basis of the MMPI, the authors argued that patients with spasmodic dysphonia had to be a remarkably heterogeneous group.

Our second aim was to determine whether symptoms of negative affectivity (depression and anxiety) and somatic complaints were consequences of the disease. Research on patients with physical symptoms or disorders underlines the changeability of psychological and somatic complaints in relation to medical treatment.²⁴ In 1991, Cannito⁸ found elevated scores of depression and anxiety among 18 patients with spasmodic dysphonia compared to matched normal controls. In addition, the patients showed abnormally elevated levels of somatic complaints. Whether these differences could be explained on the basis of a reaction to a chronic disorder or were related to a possible psychogenic origin of spasmodic dysphonia remained unsolved.

Before intervention, we also found statistically significant elevated levels of psychological and somatic complaints. These complaints decreased systematically during the course of the treatment. Compared

TABLE 5. POSTTREATMENT SCORES ON HSCL²¹ AND NORMATIVE DATA

	Spasmodic Dysphonia Group (n = 46)	Dutch Norm Group (n = 406)	<i>p</i> *
HSCL-Psych	6.1 ± 6.4	7.9 ± 6.2	.06
HSCL-Somat	2.2 ± 2.6	2.1 ± 2.3	.60
HSCL-Total	18.6 ± 18.4	22.3 ± 16.5	.18
Data are mean	± SD.		
*None of scales	differed significant	ily; 1-sample t-test, d	f = 45.

to the pretreatment scores of the HSCL, a significant reduction was found after treatment (Table 5). Moreover, the scores all reached levels within the normal range. The subjective and objective ratings of voice quality also improved systematically during treatment, establishing a normal to near-normal voice. These findings suggest that the higher pretreatment scores of psychological and somatic complaints were secondary to the voice disorder.

In just 2 studies has the effect of treatment of spasmodic dysphonia on standardized psychometric test results been assessed.^{9,10} In the first study, Murry et al⁹ demonstrated that 32 subjects displayed significantly elevated depression and anxiety in the absence of significantly elevated somatic complaints. The use of Botox had generally reduced the measures of depression and anxiety at 1 week after injection, and the effect was maintained during the ensuing 2 months. The authors concluded that the elevated depression and anxiety could have been the result of acquiring spasmodic dysphonia. These results are in line with ours, but in their study there was a problem in terms of subject compliance. They started with 32 patients, but the long-term influence (defined as 2 months after treatment) could only be determined for 13 of the original 32 subjects, so the study's validity was reduced.

The second study in which patients with spasmodic dysphonia were examined by means of psychometric tests was conducted by the same group.¹⁰ They examined the communication attitudes of 20 patients with AdSD before and after Botox injection. Although significant attitudinal improvement was observed, the patient group remained significantly different in their attitudes toward communication as compared to the normal controls. The amount of change in communication attitudes was significantly related to the amount of change in standardized psychological measures of depression and anxiety. However, this study encountered the same problem as the other study.9 Initially, 20 patients were included and could be analyzed 1 week after Botox treatment. However, only 14 of the 20 patients returned for the 2-month postinjection follow-up examination. Moreover, before the Botox injection, 6 of the 20 patients' test scores (Erickson Scale of Communication Attitudes) already fell within the normal range.

We have to emphasize that on the basis of the results of our study, no inferences can be made about causality and the role of psychological risk factors in the development of spasmodic dysphonia. However, results of recent research in the area of health psychology demonstrate that personality characteristics are stable constructs that are unrelated to somatic disorders.²⁵ On the other hand, the response to a somatic treatment does not necessarily rule out a psychogenic contribution to either symptoms or symptom severity. A longitudinal study would be more appropriate for elucidating a possible causal relation-

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ship between specific personality attributes and the occurrence of AdSD.

CONCLUSION

In this study, standardized psychometric tests were used to quantify psychological aspects of patients with AdSD. No differences in personality characteristics between AdSD patients and a representative norm group were found. However, the patients showed significantly more psychological and somatic complaints. After establishment of a (near) normal voice with Botox injections, these features were reduced to normal levels. This finding suggests that the psychological and somatic complaints are secondary to the voice disorder. These findings, and the normal personality characteristics, do not support a psychogenic cause of AdSD.

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