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Research Paper

Anxiety and depression in spasmodic dysphonia patients

Amanda Hu [a,*], Al Hillel [b], Wei Zhao [c], Tanya Meyer [b]

[a] *Division of Otolaryngology — Head & Neck Surgery, Department of Surgery, University of British Columbia, Vancouver, Canada*
[b] *Department of Otolaryngology — Head & Neck Surgery, University of Washington, Seattle, WA, USA*
[c] *Albert Einstein College of Medicine, Bronx, NY, USA*

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Abstract *Objective/Hypothesis:* Experts used to believe that spasmodic dysphonia (SD) was a psychogenic disorder. Although SD is now established as a neurological disorder, the rates of co-morbid anxiety and depression range from 7.1% to 62%. Our objective was to study the prevalence and risk factors associated with these mood disorders in SD patients.
Study design: Retrospective.
Methods: SD patients who presented for botulinum toxin injections were recruited. Demographic data, Hospital Anxiety and Depression Scale (HADS), Voice Handicap Index-10 (VHI-10), General Self-Efficacy scale (GSES), Disease Specific Self-Efficacy in Spasmodic Dysphonia scale (DSSE), and Consensus Auditory Perceptual Evaluation of Voice (CAPE-V) were collected.
Results: One hundred and forty two patients (age (59.2 ± 13.6) years, 25.4% male) had VHI-10 of 26.3 ± 6.9 (mean ± standard deviation), GSES 33.2 ± 5.8, CAPE-V 43.9 ± 20.9, HADS anxiety 6.7 ± 3.7, and HADS depression 3.6 ± 2.8. About 19 (13.4%) and 4 (2.8%) had symptoms of anxiety and depression respectively. Final linear regression model for HADS anxiety (R^2 = 32.90%) showed that patients who were less likely to have anxiety symptoms were older age ($p < 0.001$), male ($p = 0.002$), have higher GSES ($p < 0.001$) and lower VHI-10 ($p = 0.004$). Final linear regression model for HADS depression score (R^2 = 34.42%) showed that patients who were less likely to have depressive symptoms had high DSSES ($p < 0.001$).
Conclusions: Prevalence of anxiety (13.4%) and depression (2.8%) in SD were lower than previously reported in the literature. Risk factors for anxiety were: younger age, female gender,

* Corresponding author. Division of Otolaryngology — Head & Neck Surgery, Department of Surgery, University of British Columbia, Vancouver General Hospital, 4th Floor, 2775 Laurel Street, Vancouver, V5Z 4A6, British Columbia, Canada. Fax: +1 604 875 5382.
E-mail address: amanda.hu@vch.ca (A. Hu).
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lower general self-efficacy, and higher perceived vocal handicap. The main risk factor for depression was lower disease specific self-efficacy.

Introduction

In the past, spasmodic dysphonia (SD) was believed to be a psychosomatic disorder.[1–3] Experts thought that SD was a functional disorder triggered by stress, anxiety, and depression. SD was characterized as a personality disorder treated with psychotherapy.[2] It was not until the 1960s that researchers like G. Paul Moore started to discover the biological basis of SD.[3,4]

It is now accepted that SD is a focal neurological dystonia of the intrinsic muscles of the larynx.[5,6] Involuntary muscle contractions occur during speech, causing a characteristic vocal output. SD is task-specific, so the other laryngeal functions of swallowing and breathing are spared. There are three main types of SD: (1) When only the adductor muscles are affected (thyroarytenoid, lateral cricoarytenoid, and interarytenoid muscles), the patient has adductor SD with a strangled vocal quality. (2) When only the abductor muscles are affected (posterior cricoarytenoid muscles), the patient has abductor SD with a breathy vocal quality. (3) When both the adductor and abductor muscles are affected, the patient has mixed SD. The National Spasmodic Dysphonia Association estimates that approximately 50,000 people in North America are affected by SD.[5] One study from Iceland estimated the prevalence of primary laryngeal dystonia to be 5.9 per 100,000.[7] Accurate worldwide statistics are not available.[8]

Although SD is now recognized as a neurological disorder, the pathogenesis of this voice disorder is still unknown. Due to historic misconceptions, these patients may be misdiagnosed as having a psychiatric or functional disorder. To further complicate the situation, SD patients may have co-existent psychiatric disorders like anxiety and depression. Previous studies have reported co-morbid rates of anxiety and depression in SD patients to be 7.1%–62.0%.[3,9–11] This is obviously a wide range.

It is important for otolaryngologists to properly diagnose SD and to identify patientswho are at risk for mood disorders like anxiety and depression. Identifying these patients is the first step to helping them seek treatment. The objective of this study was to determine the prevalence and risk factors associated with anxiety and depression in SD patients.

Methods

Approval was obtained from the institutional review board at the University of Washington. A retrospective cohort study was conducted of all adult SD patients who presented for botulinum toxin injections from September 2011 to June 2012. Patients were excluded if they did not have the mental capacity to complete the study, if they were $<$18 years old, or if they declined to participate. Mental capacity was determined by the ENT clinician who had a long term relationship with the patient. Clinical diagnosis of dementia and inability to orient to person, place, or time was used. The Hospital Anxiety and Depression Scale (HADS) was used as a screening tool for these mood disorders.[12] A database was created with the following variables: age, gender, professional voice use, employment status, Voice Handicap Index-10 (VHI-10),[13] General Self Efficacy scale (SE),[14] and Consensus Auditory Perceptual Evaluation of Voice (CAPE-V).[15]

Hospital Anxiety and Depression Scale (HADS)[12]

HADS is a validated, reliable, screening tool for anxiety and depression in an outpatient population. There are seven questions on anxiety symptoms and seven questions on depressive symptoms experienced in the past week by the patient. Questions are scored on a four-point scale (0–3). Scores on each subscale range from 0 to 21. Normal is a score of 0–7, highly suggestive of a mood disorder is 8–10, and probable presence of a mood disorder is a score of $\geq$11. A review of the literature has reported good internal consistency for both subscales: Cronbach's α of HADS-A was 0.83 (0.68–0.93) and of HADS-D was 0.82 (0.67–0.90).[16] HADS has been used in other studies of mood disorders in voice patients. HADS has been used as the primary outcome measure for other voice studies.[17–19]

Other measurement tools

A patient's perceived handicap from his/her voice was measured with VHI-10.[13] This reliable, validated questionnaire includes 10 items answered on a scale of 0–4. Scores range from 0 to 40 and an abnormal score is above 11. VHI-10 was adapted from a longer 30 item questionnaire and has been used widely in the voice literature.

An expert clinician's evaluation of the quality of the patient's voice was measured with CAPE-V.[15] This standardized measurement tool was developed by the American Speech-Language-Hearing Association. Six vocal qualities are evaluated: roughness, breathiness, strain, pitch, loudness, and overall quality. Each attribute is measured on a visual analog scale of 100 mm by indicating the perceived deviance from normal. A higher score indicates a lower quality of voice. The CAPE-V assessments were all performed by a single speech language pathologist with over 30 years of experience in neurolaryngology and voice disorders.

The General Self-Efficacy Scale (GSES) is a validated tool used to measure self-efficacy.[14] Self-efficacy (SE) is the

psychological concept of a person's ability to cope with adversity and adapt to a new and stressful situation.[20] Self-esteem and locus of control overlap with self-efficacy. Previous studies have shown that SE is a strong predictor of health behaviors.[21–26] SE affects rates of smoking cessation[21] and adherence to therapy for chronic diseases, like diabetes.[22] SE has also been studied in various voice disorders.[23–26] The GSES is a 10 question scale where patients rate statements from 1 to 4. Total scores range from 10 to 40 and higher scores indicate higher degrees of SE. Although there is no abnormal cut-off score, a previous study of 1594 healthy American adults reported a mean GSES score of 29.48 ± 5.13.[27]

Disease Specific Self-Efficacy in Spasmodic Dysphonia scale (DSSES) was created by our group.[26] We felt that SD was such a unique disorder that it warranted the creation of its' own scale. Patients with this chronic voice disorder adapt well over time and become empowered with their knowledge and experience with their condition. SD patients become active participants in their health care. The DSSES includes a total of 13 questions: 8 questions from the GSES and 5 disease-specific questions. There is also no abnormal cut-off score and a higher score indicates a higher degree of disease-specific SE. A previous study of SD patients treated with botulinum toxin injections reported a mean and standard deviation of 42.1 ± 6.9 out of 52.[26]

Statistical analysis

Descriptive statistics, including measures of central tendency, were used to characterize the study population. Multivariate analyses with logistic regression models were conducted in R (version 2.15.2). In multivariate analysis, model selection procedures based on Akaike Information Criteria (AIC) were performed to identify the most informative yet parsimonious linear regression models.

Results

One hundred and forty two patients completed the study. Five patients were excluded: one patient declined to participate, one patient had dementia, and three patients failed to complete all the forms. Table 1 shows the demographic data of the study population. The mean age was 59.2 years with a range of 18–87. About 25.4% were male and 95.8% had adductor SD.

To investigate the factors associated with the HADS-A and HADS-D scales, linear regression on these two outcome measures were performed respectively. In the analyses with respect to both of the scores, a full model which incorporated all possible factors, a model selected by a model-selection procedure based on AIC and a reduced model comprised only those significant factors in the AIC-selected model were constructed and evaluated. An F-test was performed to test whether the factors removed from the AIC-selected model had statistically significant contribution to the model. Goodness-of-fit statistics R^2 and adjusted R^2 were also reported for the three models.

The factors that are associated with HADS-A in the full model are age, gender, GSES, VHI-10, and CAPE-V pitch (Table 2). Model-selection procedure based on AIC identified a model which comprised the same five significant factors in the full model as the best model. We further removed the non-significant CAPE-V pitch in the AIC-selected model and derived the reduced model. An F-test showed that the reduced model was not statistically significantly different from the AIC-selected model ($p = 0.110$). The final model, which was the most informative yet parsimonious and explained 32.90% of the variance in the dependent variable, indicated that older age ($p < 0.001$), male ($p = 0.002$), a higher GSES score ($p < 0.001$) and a lower VHI score ($p = 0.004$) were associated with a lower HADS-A. The effect sizes of the four factors on HADS-A were consistent across the three models, which indicated a lack of confounders in the factors available. Gender had a large effect on HADS-A. Men on average were two points lower than women on this score.

In the full model regarding HADS-D, DSSES was the only factor that was statistically significantly associated with HADS-D (Table 3). The model selection procedure based on AIC selected a model which comprised age, employment, DSSES and VHI-10. The reduced model, which only included DSSES, was not statistically significantly different from the AIC-selected model (F-test, $p = 0.114$). The reduced model indicated that a one-unit increase on DSSES on average was associated with a 0.391 decrease on HADS-D ($p < 0.001$). This effect size and its direction of DSSES on HADS-D were largely consistent across the three models. We also noticed that the reduced model (adjusted $R^2 = 34.42\%$), which only has DSSES as the independent variable but explained 34.91% of the variance in HADS-D, was not inferior than the full (adjusted $R^2 = 35.91\%$) or the AIC-selected (adjusted $R^2 = 33.38\%$) models.

Table 1 Demographic data of the study population ($n = 142$).

Age (mean ± standard deviation) (years)	59.2 ± 13.6
Male (n (%))	36 (25.4%)
Adductor (n (%))	136 (95.8%)
Professional Voice User (n (%))	24 (16.9%)
Employed (n (%))	81 (57.0%)
Voice Handicap Index – 10 (mean ± standard deviation)	26.3 ± 6.9
Consensus Auditory Perceptual Evaluation of Voice – Overall (mean ± standard deviation)	43.9 ± 20.9
General Self-Efficacy Score (mean ± standard deviation)	33.2 ± 5.8
Disease Specific Self-Efficacy in Spasmodic Dysphonia Scale (mean ± standard deviation)	32.8 ± 5.1
Hospital Anxiety and Depression Scale – Anxiety (mean ± standard deviation)	6.7 ± 3.8
Hospital Anxiety and Depression Scale – Depression (mean ± standard deviation)	3.6 ± 2.8

Table 2 The linear regression results on Hospital Anxiety and Depression Scale (HADS) - anxiety score.

Variable	Full model			Selected model based on AIC			Reduced model		
	Effect	SE	*p*-value	Effect	SE	*p*-value	Effect	SE	*p*-value
Age	−0.080	0.023	<0.001	−0.076	0.020	<0.001	−0.076	0.020	<0.001
Gender	−1.630	0.646	0.013	−1.854	0.625	0.004	−1.939	0.627	0.002
GSES	−0.190	0.082	0.022	−0.283	0.051	<0.001	−0.291	0.051	<0.001
VHI-10	0.131	0.041	0.002	0.120	0.040	0.003	0.119	0.040	0.004
CAPEV-P	0.033	0.017	0.050	0.024	0.015	0.110			
CAPEV-O	0.024	0.025	0.333						
CAPEV-R	0.002	0.015	0.899						
CAPEV-B	−0.030	0.032	0.357						
CAPEV-S	−0.034	0.020	0.087						
CAPEV-L	0.012	0.023	0.592						
SD Type	1.562	1.502	0.301						
DSSES	−0.119	0.100	0.236						
Employment	−0.167	0.654	0.799						
R^2	39.75%			36.19%			32.90%		
adjusted R^2	33.28%			33.71%			32.90%		

GSES: General Self Efficacy scale.
VHI−10: Voice handicap index 10.
CAPEV-P: Consensus Auditory Perceptual Evaluation of Voice − pitch.
CAPEV-O: Consensus Auditory Perceptual Evaluation of Voice − overall.
CAPEV-R: Consensus Auditory Perceptual Evaluation of Voice − roughness.
CAPEV-B: Consensus Auditory Perceptual Evaluation of Voice − breathiness.
CAPEV-S: Consensus Auditory Perceptual Evaluation of Voice − strain.
CAPEV-L: Consensus Auditory Perceptual Evaluation of Voice − loudness.
SD: Spasmodic dysphonia.
DSES: Disease Specific Self-Efficacy in Spasmodic Dysphonia scale.

Table 3 The linear regression results on Hospital Anxiety and Depression Scale -Depression score.

Variable	Full model			Selected model based on AIC			Reduced model		
	Effect	SE	*p*-value	Effect	SE	*p*-value	Effect	SE	*p*-value
DSSES	−0.291	0.075	<0.001	−0.366	0.047	<0.001	−0.391	0.046	<0.001
Age	−0.031	0.018	0.084	−0.029	0.017	0.083			
VHI-10	0.048	0.031	0.128	0.046	0.029	0.118			
Employment	−0.656	0.493	0.186	−0.768	0.461	0.098			
CAPEV-O	−0.013	0.019	0.503						
CAPEV-R	0.012	0.011	0.306						
CAPEV-B	0.000	0.024	0.996						
CAPEV-S	−0.003	0.015	0.823						
CAPEV-P	−0.005	0.013	0.712						
CAPEV-L	0.018	0.017	0.298						
Gender	−0.327	0.487	0.503						
SD Type	0.271	1.133	0.811						
GSES	−0.074	0.062	0.232						
R^2	39.85%			37.82%			34.91%		
adjusted R^2	33.38%			35.91%			34.42%		

DSES: Disease Specific Self-Efficacy in Spasmodic Dysphonia scale.
VHI−10: Voice handicap index 10.
CAPEV-O: Consensus uditory Perceptual Evaluation of Voice − overall.
CAPEV-R: Consensus uditory Perceptual Evaluation of Voice − roughness.
CAPEV-B: Consensus Auditory Perceptual Evaluation of Voice − breathiness.
CAPEV-S: Consensus Auditory Perceptual Evaluation of Voice − strain.
CAPEV-P: Consensus Auditory Perceptual Evaluation of Voice − pitch.
CAPEV-L: Consensus Auditory Perceptual Evaluation of Voice − loudness.
SD: Spasmodic dysphonia.
GSES: General Self Efficacy scale.

Discussion

The previously reported co-morbid rates of anxiety and depression in SD patients span a wide range: 7.1%–62.0%.[3,9–11,28] There are several reasons for this wide range. First, different studies used different methods to measure anxiety and depression. Some studies used a previous psychiatric diagnosis,[3] other studies used a structured psychiatric interview,[9,11,28] and other studies used standardized questionnaires.[9–11] Second, the sample sizes from the previous studies varied widely, from 10 to 127.[3,9–11,28] Lastly, studies published in different time periods may have been biased by the prevailing beliefs of the era. A review of the literature provides historical perspective on how clinicians studied mood disorders in the spasmodic dysphonia patient population.

One of the earliest studies was by Aronson et al[11] in 1968. They conducted clinical interviews with 29 SD patients and used the Minnesota Multiphasic Personality Inventory. They concluded that 62% of patients showed psychiatric symptoms. This study was published when SD was still believed to be a psychogenic disorder. This study also quoted the highest prevalence among all the studies in our literature review.

Liu et al[9] conducted a study in 1998 with 10 SD patients and 20 controls. A psychiatrist interviewed the patients and administered psychometric testing, including the Hamilton Depression Rating Scale (HDRS) and Hamilton Anxiety Rating Scale (HARS). SD patients scored higher on the subscales of somatization, obsessive-compulsive symptoms, depression, anxiety, and psychoticism than the normal controls. Patients were evaluated before botulinum toxin injection and 1 month after the procedure. The scores on the psychometric testing all improved after the botulinum toxin injection. They concluded that the mood symptoms of SD patients were secondary to the voice disorder and not the etiology of the disorder.

In 2003, Mirza et al[10] used the Brief Symptom Inventory to measure "psychiatric caseness" (i.e. a clinically significant psychiatric distress that is indicative of a psychiatric disorder). Among the 17 SD patients, 1/17 (7.1%) tested positive for a major psychiatric disorder. This study's results may have been affected by the small sample size.

In 2007, Gundel et al[28] from Germany interviewed 50 SD patients and 27 patients with vocal fold paralysis with the Structured Clinical Interview for DSM-IV (SCID-I). They reported a 41.7% rate of comorbid psychiatric disease, which was significantly higher than the control group of 19.5%.

In the most recent paper in 2012, White et al[3] from the Emory group performed a case cohort study with 128 SD patients and 146 patients with benign vocal disorders. They analyzed the number of these patients who already had a diagnosis of anxiety and depression. They reported that among the SD patients, 28.3% had depression and 25.2% had anxiety. There were no significant differences in the prevalence of these mood disorders in SD and other benign vocal disorders. This study represented a treatment-seeking population — patients whose psychiatric symptoms were severe enough to seek medical attention. The results may have under-reported patients with mild symptoms who have not yet been diagnosed.

The current study used the Hospital Anxiety and Depression Scale (HADS). This measurement tool was chosen because previous studies have reported good internal consistency for both subscales[16] and it has been used in previous studies of voice disorders.[17–19] For example, Dietrich et al[17] from the University of Pittsburgh group used HADS to study anxiety and depression with voice patients with paradoxical vocal fold movement disorder, muscle tension dysphonia, benign vocal fold lesions, and glottic insufficiency. The reported prevalence of anxiety and depression ranged from 25.0 to 35.6% with these voice disorders.

The current study reported a 13.4% prevalence of anxiety and 2.8% prevalence of depression. The National Institutes of Health estimated that the prevalence of anxiety and depression among a general population of US adults were 18.1% and 6.7% respectively.[29–30] Both results are lower than the general population. The prevalence of anxiety is within the reported range in previous voice disorders, but the prevalence of depression is lower than the range reported in previous voice disorders.

There may be an explanation for this low level of anxiety in the current study. The SD patients were recruited from a mature laryngology practice with a mean duration of disease of over a decade. These SD patients were all well-established on their botulinum toxin doses. SD patients may experience more symptoms of anxiety earlier on in their disease. Most SD patients have a frustrating course early on before they are finally diagnosed. The first few botulinum toxin injections can also represent an adjustment period.

Female gender has been reported in previous studies to be a risk factor for depression and anxiety in the general population and in voice disorder patients.[3,17,27,28] Our study confirmed that gender was a risk factor for anxiety in SD patients.

Younger age has been reported in previous studies to be a risk factor for depression and anxiety in the general population and in voice disorder patients.[3,30,31] Our study confirmed that younger age was a risk factor for anxiety in SD patients.

Expert clinical judgement of the severity of vocal impairment in SD was reported by Gundel et al[28] to be positively associated with the presence of a mood disorder. Seven voice professionals used the Unified Spasmodic Dysphonia Rating Scale[32] to blindly judge the quality of the patients' voices. The strained–strangled voice quality was in particular found to be significant. The current study used CAPE-V as a measure of an expert clinician's judgement on the quality of the patients' voice. This variable was not significant in our current study. In contrast, a higher VHI score was associated an increased risk of anxiety. VHI is different than CAPE-V and the Unified Spasmodic Dysphonia Rating Scale in that it is a patient-administered questionnaire that measures self-perceived vocal handicap. Several reasons may explain these divergent results. First, different measurement tools were used in the two studies. Second, our study had a larger sample size ($n = 142$) than the Gundel at et study ($n = 50$). Lastly, the clinician's perceptual judgement of the quality of a patient's voice may be different than the patient's self-perception.

The patient's subjective assessment of "satisfaction with health" was reported by Gundel et al[28] to be negatively

associated with the presence of a mood disorder. The previous study used Questions on Life Satisfaction (FLZM),[33] a short questionnaire on general and health related quality of life, to measure this variable. The present study used the GSES and DSSES. The latter measurement tool was created by our group to measure self-efficacy in the SD population.[26] The current study reported that DSSES was the only independent variable in the regression model of depressive symptoms. Since the psychological concept of self-efficacy includes concepts of self-confidence, self-esteem, and the ability to cope with adversity, it was not a surprise that this was an important risk factor for depression. Self-efficacy has also been associated with depression in other chronic diseases, like hemodialysis,[34] diabetes,[35] rheumatic disease,[36] and pregnancy.[37]

There are some limitations to our study. First, the linear regression models only explained about 35% of the variance observed in the anxiety and depression scores; thus, there may be other important factors that were not accounted for. For example, a family history of psychiatric disorders and substance abuse were not specifically elicited in the histories. This leads to our second limitation: this was a retrospectives study. Some questions that are usually less important in the ENT history, like a family history of mood disorders, were not specifically asked. Lastly, we did not have a psychiatrist perform clinical interviews with our patients. Due to human resource and time limitations, this option was not feasible.

Conclusions

The prevalence of anxiety (13.4%) and depression (2.8%) in SD patients in our study were lower than previously reported in the literature. Risk factors for anxiety were: younger age, female gender, lower general self-efficacy, and higher perceived vocal handicap. The main risk factor for depression was lower disease specific self-efficacy. Clinicians can use this information to identify voice patients who are at risk of developing these mood disorders.

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