

A Preliminary Investigation of the Affective, Behavioral and Cognitive Variables Associated with Spasmodic Dysphonia

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Abstract

Background: Spasmodic dysphonia (SD) is often assessed and treated in a rather mono-dimensional way, despite knowledge of the impact of SD on a person's quality of life. This study investigates how adults with SD think and feel about their speech, and cope with their voice problem.

Methods: Adults with SD were asked to fill out the Behavior Assessment Battery's (BAB-Voice) self-report tests. The Speech Situation Checklist-Emotional Reaction and Speech Disruption investigate negative emotional reaction and voice problems in particular speech situations. The Behavior Checklist provides an inventory of behaviours that might be used to avoid or escape voice problems. The Communication Attitude Test for Adults assesses speech-associated attitude.

Results: Adults with SD had elevated scores on all BAB tests. Their results were comparable to BAB research with people who stutter.

Conclusion: The data show the BAB's potential to assist clinicians in diagnosis and in designing a tailored multi-dimensional treatment plan.

Keywords: Spasmodic dysphonia; Behavior Assessment Battery; Multi-dimensional assessment; Evidence-based assessment; Stuttering

Background

Spasmodic dysphonia (SD) is a task specific, focal dystonia characterized by involuntary spasms of the laryngeal muscles. SD is typically an adult-onset disorder [1] that is more prevalent in women (nearly 80%) than in men [2]. The National Spasmodic Dysphonia Association estimates that 50,000 people in North America are affected by SD. The literature provides evidence in support of a neurogenic etiology [3-12] more recently linking SD to the basal ganglia, thalamus, sensorimotor cortex and cerebellum [13,14]. In the past, SD has been associated with controversy in the literature as it relates to varied neurologic, psychiatric and psychoneurotic variables [14-19]. Whereas the theory that SD is attributed to a psychogenic disorder has been disproven, it is evident that a high proportion of patients with SD experience emotional reactions/disturbances, especially anxiety and depression [20].

SD can present in a variety of forms (adductor, abductor, tremor and mixed), each exhibiting distinct vocal symptoms. Adductor SD (ADSD) has been considered the most common form of SD and is characterized by irregular tight or strained-strangled voice stoppages that are caused by the intermittent hyper-adduction of the vocal folds during phonation [21].

'Speech interruption,' a characteristic fluency break commonly associated with stuttering is frequently experienced by individuals with ADSD due to irregular muscular spasms. As with stuttering,

individuals with ADSD may also demonstrate increased vocal effort during speech tasks, particularly when attempting to push through a spasm. As such, laryngeal involvement has been evidenced during these interruptions in the forward flow of speech [21,22].

The fact that people who stutter (PWS) exhibit more than just stuttering behaviours has been made clear in repeated research investigations [23-27]. Aside from an interruption in the forward flow of speech, PWS might use behaviours that are secondary to their stuttering, have a negative speech-associated attitude and experience negative emotional reaction related to sounds, words and/or situations. These dimensions provide clinicians with a multi-dimensional "view from within" [27] of their clients' difficulties.

Over the past decades, the Behavior Assessment Battery (BAB) [24,25,27,28] has been internationally used to investigate the aforementioned dimensions among children and adults who stutter. The BAB consists of a battery of self-report tests: the Speech Situation Checklist (SSC), the Behavior Checklist (BCL) and the Communication Attitude Test for Adults (BigCAT). The BAB provides norms for PWS and people who do not stutter (PWNS). It aids in differential diagnosis of people with and without fluency disorders.

In addition, the standardized tests that make up the BAB assist in determining the best treatment options for PWS. They point to components that need to be part of the treatment plan, such as addressing a person's negative thoughts about his or her speech, dealing with the coping behaviours being employed, and attend to the difficult situations that create anxiety and evoke speech problems.

Historically, psychosocial implications of SD have received little attention in the research literature [29]. However, over the past decades investigations have explored the psychosocial impact of SD on daily life by utilizing scales of patient perception developed for a wide range of voice disorders [30-33]. The intent of utilizing such scales is to emphasize the meaning of impairment from the patient's perspective, so that interventions have the potential to produce the greatest benefit [34], however, these scales are not specific to SD. In this vein, scales such as the Voice Handicap Index (VHI) [35,36], Voice-Related Quality of Life (V-RQOL) [37,38], and the Voice Disability Coping Questionnaire [39] have been used as a widely-reported means of measuring the psychosocial impact of voice disorders.

Attempts to understand the psychosocial effect of SD has also been reported in the literature through the use of more general health-related questionnaires [19,40-42]. While not specifically focusing on SD, they attempt to measure the use of coping behaviours within the social and occupational setting.

Though the aforementioned work has broadened our understanding of the psychosocial consequences of SD, much is still unknown about the situational difficulties, speech-related anxiety, attitude and coping strategies used by individuals with SD. Gaining an authentic view of what goes on within an individual suffering from SD continues to be a challenge. This *inside view* can be difficult to obtain because of the unique and intricate manner in which a voice disorder interacts with many personal and environmental variables that shape each individual's experience [29].

While SD has been consistently reported to restrict individuals from social and occupational participation; the environments, the coping mechanisms and the personal factors that shape the consequences of individuals living with SD require further exploration [29]. All too often, SD is still assessed and dealt with in a rather mono-dimensional way, although clinical experience has made it clear that individuals with SD suffer from more than just a voice/speech problem.

Given the perceived similarities between stuttering and SD and the fact that, aside from research that relates to quality of life, little is known about the aforementioned variables that surround SD, it was the purpose of this pilot study to assess the behaviours of avoidance and escape, speech-associated attitude, emotional reaction and speech disruption related to particular situations, among individuals with SD.

In order to determine the extent to which individuals with SD report using coping behaviours to deal with their speech problem, think negatively about their speech, and experience difficult speech situations, an adaptation of the existing BAB was administered to individuals with SD.

Methods

Participants

The Behavior Assessment Battery (BAB) – Voice [43] was administered to 41 individuals with SD. Thirty-two of the participants were female, nine were male. Their ages ranged from 35 to 81 years, with a mean and median age of 60 years. Twenty five percent were under age 50, 75% were under age 68. The participants were drawn from attendees of the 2011 National Spasmodic Dysphonia Association's (NSDA) conference and came from different geographical areas within the United States of America.

Materials and procedure

Each of the participants was instructed to complete the different BAB self-report tests on an individual base. The NSDA attendees were given the tests by graduate students who were trained by the first author, who is also the BAB co-author, in the administration of the test battery. The order in which the tests were given to the participants was randomized to guard against an order effect.

The four different BAB self-report tests were adapted to be more appropriate for individuals with SD disorders. In other words, the test instructions and items were written in such a way to be specific to SD and to reflect the impact of SD on a person's speech-associated belief, the possible use of coping behaviours and the extent to which anxiety and voice difficulty is linked to particular speech situations. This adaptation resulted in the BAB-Voice [43].

The Communication Attitude Test for Adults (BigCAT) investigates a person's speech-associated attitude. In other words, it tests what an individual thinks about his or her voice and way of speaking. The participants are asked to reflect on the 35 BigCAT statements and to indicate whether or not a statement indicates how they perceive their voice. They do this by circling 'true or false' to assertions such as "There is something wrong with my voice; my voice limits my future". This dichotomous scale uses a zero – one score for each item, whereby a score of 1 represents a negative speech-associated attitude. Thus, the possible score for the BigCAT can range from 0 to 35.

The Speech Situation Checklist (SSC) consists of two sections. SSC-Emotional Reaction (SSC-ER) assesses negative emotional reaction (anxiety, fear, worry) in particular speech situations. The second section, SSC-Speech Disruption (SSC-SD) is designed to determine the extent of a person's speech disruption in the same 51 situations as described in SSC-ER. Examples of speech situations referred to are: "talking on the telephone; being interviewed for a job". The SSC uses a Likert-type scale whereby the participants indicate, on a 5-point scale, the extent to which each situation creates negative emotion (SSC-ER) or speech disruption (SSC-SD). The scores are summed and can range from 51 to 255.

The Behavior Checklist (BCL) investigates the use of coping behaviours. In column I, the participant marks those behaviours used as avoidance or escape behaviours in an attempt to deal with the voice problem. Ninety-five different possible coping behaviours are described. They range from movement of body parts, to behaviours more directly linked to the act of speaking such as the use of interjections and word substitutions, changes in voice intensity and intonation, etc. To obtain a total score on the BCL, the behaviours checked-off are summed. The scores can range from 0 to 95. In column II, the frequency with which the marked behaviours are being used is indicated on a five-point scale ranging from 'very infrequently' to 'very frequently'.

Results

Statistical analyses were performed using IBM SPSS Statistics 22 software. As indicated in Table 1, the BigCAT yielded an average score of 30.36 (SD=5.97), with a range from 4 to 35 (out of a possible maximum of 35). The median and mode were 32.5 and 34, respectively.

	BigCAT	SSC-ER	SSC-SD	BCL
Mean	30.36	162.48	164.65	16.57
Standard Deviation	5.97	45.83	46.50	9.73
Median	32.50	171.50	175.00	16.00
Mode	34	189	175	22
Min	4	63	58	0
Max	35	247	251	42

Table 1: Measures of central tendency and variation for each of the BAB sub-tests.

The mean score for SSC-ER was 162.48 (SD=45.83) with scores ranging from 63 to 247 possible maximum is 255). The median score obtained was 171.50 and the mode was 189. The average score on SSC-SD was 164.65 (SD=46.50). The scores ranged from 58 to 251. The median of 175.00 was identical to the mode.

Finally, the average BCL score was 16.57 (SD=9.73). The scores ranged from 0 to 42 (out of a possible maximum of 95). The median BCL score was 16.00 and the mode was 22.

Of special interest was the relationship between the two sections of the SSC. Comparison of the two test means, using t-tests, revealed that they did not differ significantly ($t=0.988$; $p=0.329$). In addition, Pearson Product Moment correlations indicated that SSC-ER and SSC-SD correlate highly ($r=.96$, $p=0.05$). In addition, the fact that the different sub-tests that compose the BAB are related to one another was exemplified by significant correlations. The BigCAT correlated 0.69 and 0.70 ($p=0.000$) with SSC-ER and SSC-SD, respectively. The lowest correlations were between the BCL and BigCAT ($r=0.56$; $p=0.000$), SSC-ER ($r=0.42$, $p=0.011$) and SSC-SD ($r=0.40$, $p=0.014$).

Discussion

A major aim of this pilot study was the determination whether or not individuals with SD show elevated scores on the subtests of the BAB, as has been seen among PWS. This was indeed found to be the case. As a group, the individuals with SD in this investigation had raised scores on all of the BAB tests.

As far as the BigCAT is concerned, compared to data obtained by Vanryckeghem and Brutten [27], PWS scored an average of 26.68 (SD=5.33), a score that was significantly higher compared to that of PWNS (mean=3.84 SD=3.67). Although it needs to be kept in mind that an adaptation from the original BigCAT was employed in the current investigation, the scores of the SD participants (mean=30.36) closely resembled those of PWS (<1 SD above the PWS mean). In addition, the scores of individuals with SD clearly fall several standard deviations above the scores obtained on 'typical speakers' in the 2011 study. Clearly, the speech-associated attitude of individuals with SD can be determined as "negative". This finding is in agreement with past literature investigating the consequences of SD on communication-related quality of life, indicating that the presence of ADSD related to frustration, embarrassment, self-consciousness, and feelings of incompetence [29].

The current SSC test scores are similar to data obtained with PWS in Belgium [24]. Again, keeping in mind that the current SSC forms

are adaptations of the SSC for PWS, the current SSC-ER data (162.48) are essentially double those obtained for PWNS in that study (82.54) and higher than those of PWS (126.88). Similarly, the SSC-SD scores for the SD participants (164.65) are substantially higher than those of PWNS (73.71) and even higher than those of PWS (130.10) in the 2003 investigation. It is clear that individuals with SD report to experience a high level of negative emotional reaction and speech disruption in particular speech situations. Examples of problematic situations are: talking on the telephone, talking to a stranger, giving your name, saying a sound or word that has previously been troublesome. These findings confirm what has recently been reported in the literature, that individuals with SD perceive their voices as not dependable. As a result, they avoid speaking out of fear of embarrassment and an inability to predict voice quality [29].

The fact that individuals with SD reports to use, on average, 16 coping behaviours, is impressive. Compared to the BCL data of PWS and PWNS [44], the SD participants scored slightly below the mean of PWS (20.79; SD=11.39), and 2 standard deviations above the mean of PWNS (5.38; SD=5.56). These data indicate that individuals with SD reported using an elevated number of avoidance and escape behaviours in order to cope with the physical manifestations of the voice disorder. Some of the more commonly reported avoidance behaviours include: avoiding eye contact, looking to the other side, pretending to be thinking about an answer, substituting one word for another, and others. These findings substantiate what has been reported by Cannito and colleagues [19,40] as communication apprehension which can lead to avoiding communication all together. Furthermore, negative thoughts related to speech performance may cause individuals with ADSD to avoid certain work environments which can, in turn, limit employment opportunities and job advancement (due to expected increase in vocal demand). For some, ADSD might lead to a complete career change, because of the fear of employability in face of the chronicity of their voice disorder. These phenomena are, once again, very similar to what is being described by PWS, who consistently report the negative ramifications of stuttering on their personal and professional life, and the fact that they use mechanisms to cope with the limitations of their speech impediment.

Conclusion

The results of this study are interesting. Never before, have individuals with SD been questioned, in a systematic way, using a standardized test, about how they think about their speech, the coping behaviours they might use, and the situations that evoke anxiety and affect their speech. While the routinely used Quality of Life scales capture general information about the impact of a voice disorder on a person's daily life, they are not specific to SD. In a non-confounding way, the introspective information obtained through the BAB gives the clinician an "inside view" [25] of the affective, behavioural and cognitive sequel of the disorder. It adds important dimensions, allows for a more fully rounded assessment of those with SD, and provides additional information to better plan a multi-modal treatment addressing an array of psychosocial components that are related to SD.

Evidently, this pilot study requires follow-up and further consideration. Similar to research investigations with PWS, a comparison of a different sample of adults with SD to a group of 'typical speakers', matched for age and gender, would shed light on whether or not the scores of SD individuals differ to a statistically significant extent from those without a voice disorder. In addition, item analysis of the different tests will allow for possible modification

of the current test forms. Clearly, given disorder incidence difference between spasmodic dysphonia and stuttering, the comparison between the members of both groups was different in terms of gender ratio. Whereas SD is clearly a female-prone disorder, the opposite is the case for stuttering. Nevertheless, studies with the BAB for PWS have pointed to the fact that gender did not affect the BAB sub-tests' scores. As such, we feel confident that the comparisons made here were not affected by the difference in gender ratio.

It is apparent that the notion among researchers and clinicians that adults with SD suffer from more than a voice problem, is a fact. Their similar trait to PWS has also been confirmed by means of this research. Another interesting elaboration of this study would be the investigation of the extent to which different voice problems have a differential effect on the BAB test scores. This, in turn, could lead to differential treatment of individuals with voice disorders.

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