Patient Perceptions of Factors Leading to Spasmodic Dysphonia: A Combined Clinical Experience of 350 Patients

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Purpose: Spasmodic dysphonia (SD) is an idiopathic voice disorder that is characterized by either a strained, strangled voice quality or a breathy voice with aphaic segments of connected speech. It has been suggested that environmental factors play a role in triggering the onset. Clinical observation suggests that some patients associate onset with specific events or factors while others do not. The purpose of this study was to examine a large database of SD patients to determine if specific triggers are associated with the onset of SD.

Procedures: Retrospective chart review.

Results: A total of 350 charts of patients with SD were identified and were categorized as either “sudden onset” or “gradual onset.” One hundred sixty-nine recalled their circumstances surrounding onset. Forty-five percent of these patients described the onset as sudden. Patient perceptions of inciting events in the sudden onset group were identified 77% of the time and 2% of the time in the gradual onset group. The most common factors identified were stress (42%), upper respiratory infection (33%), and pregnancy and parturition (10%).

Conclusions: Thirty-five percent of SD patients perceive their disorder to have a sudden onset with identified inciting events. This prevalence raises questions regarding possible behavioral and environmental factors surrounding the onset of this disorder.

Key Words: Spasmodic dysphonia, sudden onset, gradual onset.

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INTRODUCTION

Spasmodic dysphonia (SD), or focal laryngeal dystonia, is an idiopathic disorder in which voice production is impaired by irregular and uncontrolled contractions of the laryngeal muscles. Typically, it is divided into two types—adductor (~82%) and abductor (~17%)—based on the predominant effect of inappropriate muscle contraction. Adductor SD patients experience intermittent hyperadduction of the vocal folds, which results in a choked or strangled voice quality. Those with abductor SD exhibit a breathy voice quality with aphaic or whispered breaks in phonation. SD is a chronic condition, which interferes with patients’ daily communication, resulting in significant occupational disability and social isolation.

Historically, SD was considered a psychiatric condition highly related to stress, despite the conspicuous lack of success of psychiatric treatment. The early work of Dedo involving recurrent laryngeal nerve block and section demonstrated beyond a doubt that the disorder was neurologic in nature. Others have reported successful temporary relief of symptoms with botulinum toxin injections to the laryngeal musculature. Despite these therapeutic successes, knowledge regarding causes and predisposing factors remains extremely limited. To date, six “DYT” gene loci characterized by dystonia have been designated, although none have been specifically linked to SD. Additionally, no genetic studies have been reported in families with SD. Most cases occur sporadically and are not associated with other medical conditions. Women do appear more likely to be affected, but the implications of this for pathophysiology are not clear.

Some affected individuals relate the onset of their voice disorder to specific events, whereas others do not. The Laryngeal and Speech Section of the NIH has emphasized that studies that identify associations with the onset of SD provide clues for animal models, yet no large-scale study has yet examined the patient perception regarding onset of SD. The lack of a large SD population in any one center, the assessment process, and/or the lack of a consistent case history format to study the disorder may explain this. The present study aims to examine patient perceptions regarding the onset of their disorder.

MATERIALS AND METHODS

Institutional review board approval was obtained. The clinical records of patients from two urban laryngology practices...
who were evaluated and in many cases treated for SD over 2 years (2009–2010) were reviewed. Initial visit notes (some antedating the patient’s visit during the review period) were identified and reviewed, as well as accompanying neurology and speech pathology records. Patients with a diagnosis of spasmodic dysphonia of either adductor or abductor type made by one or both of the senior authors (A.B., L.S.) were included, regardless of treatment status. Details regarding the onset of spasmodic dysphonia were compiled. This was possible as the two senior authors used a similar case history format in acquiring patient data. Specifically, notation was made of the patient’s gender, age at onset of disorder, onset described as sudden or gradual, duration of onset if described as gradual (number of months or years), identification of a specific trigger associated with onset, and pertinent personal medical history including diagnoses of other focal dystonias. Sudden onset was defined as within 1 week’s time; otherwise it was considered gradual.

Patients were divided into specific subsets, including: gender, those who identified onset as sudden or gradual, and those who perceived a trigger and those who did not. A two-tailed Fisher’s exact test was performed to compare these specific subsets identified in this study (P-values < .05 were considered significant). Also, demographic information and relevant medical history were included for review.

RESULTS

Three hundred fifty patients with SD were identified. Two hundred forty (80%) of these were female and 110 (20%) were male. Seven patients (2%) reported a personal history of an additional focal dystonia (e.g., blepharospasm, writer’s cramp or torticollis) (see Table I). The mean age of onset was 46.1 ± 15.2 years. One hundred sixty-nine (59.2%) patients indicated having specific perceptions regarding the onset of their disorder and reported them at the time of their first visit. Seventy-seven (45%) of these patients with specific perceptions described the onset as “sudden” (developing within 1 week’s time). Of these 77 patients who described a sudden onset, onset-associated factors were identified 77% of the time. The remaining 92 (55%) of the 169 patients with specific perceptions regarding onset described the onset as “gradual.” The average duration of gradual onset was 6.3 ± 7.2 years. In the gradual onset group, patients associated specific factors with the onset of SD only 2% of the time (see Table II). Overall, of the factors identified, stress (n = 26 [42%]), upper respiratory infection (n = 20 [33%]), pregnancy and parturition (n = 6 [10%]), and intubation for a surgical procedure (n = 4 [6%]) were most common (see Fig. 1). Other specific factors included cessation of habitual heavy drinking (gradual), exposure to fiberglass (sudden), beginning immunotherapy (sudden), and smoking cessation (sudden). Of those who identified triggers, 52 (85.2%) were female and 9 (14.8%) were male. Sixty-nine (75%) of those in the gradual onset group were female and 23 (25%) were male. In the sudden onset group, 66 (86%) were female and 11 (14.3%) were male. Using a two-tailed Fisher’s exact test, we identified statistical significance in gender distribution when comparing the total population of SD patients studied to those who identified onset as sudden (P = .002) and to those who identified specific triggers (P = .009). Thus, we found that women were statistically more likely to have both sudden onset and identifiable triggers. Statistical significance was not found with regard to gender distribution when comparing the total population of SD patients studied to those who identified onset as gradual (P = .282). Also, in terms of gender distribution, when comparing the subsets of sudden onset versus gradual onset, no significant differences in gender were found (P = .12).

DISCUSSION

Causes of SD have been little studied even though it has been clinically observed that many patients report specific events or factors associated with onset. In 2008, Ludlow et al.6 commented that because SD may not have a strong genetic component, further investigation of potential environmental determinants might be “fruitful.” Nearly a decade ago, Schweinfurth et al.7 distributed surveys to 168 patients with SD to examine

![Frequency of perceived triggers](image-url)

**TABLE I. Study Population Characteristics.**

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<thead>
<tr>
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<th>N (%)</th>
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<tbody>
<tr>
<td>SD patients identified</td>
<td>350</td>
</tr>
<tr>
<td>Female SD patients</td>
<td>240 (80)</td>
</tr>
<tr>
<td>Male SD patients</td>
<td>110 (20)</td>
</tr>
<tr>
<td>Personal history of additional focal dystonia</td>
<td>7 (2)</td>
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SD = spasmodic dysphonia.
risk factors and demographics. An incidence of childhood measles and mumps infection of 65% of individuals was discovered. Additionally, 21% associated the onset of their disorder with psychological stress, whereas 30% associated the onset with an upper respiratory infection. Recently, Tanner et al.\(^8\) interviewed 150 patients with SD to better understand the factors associated with the disorder. Compared with a group of unaffected controls, patients with SD had an increased likelihood of a personal history of mumps, blepharospasm, tremor and intense voice use, an immediate family history of meningitis, tremor, cancer, and compulsive behavior, and an extended family history of cancer and tremor. In line with these studies, our review also identifies “stress” and “upper respiratory infection” as relevant factors frequently identified by patients. Our review also identified pregnancy and parturition, as well as endotracheal intubation as potential predisposing events. Our study did not fully examine family histories of neurologic disorders, or personal history of infectious disease as identified by Schweinfurth and Tanner. The high association of predisposing factors in the sudden onset SD group is to our knowledge a new finding in the SD population. The current data suggests that patient-related and/or environmental factors appear to be strongly related to the development of SD in a subgroup of patients who develop symptoms quickly.

SD may not be the only focal dystonia associated with specific environmental triggers. Recently, Kumar and Jog\(^8\) reported a “posttraumatic dystonia” or “syndrome” based on the premise that peripheral trauma can predispose to abnormal posturing of a specific body part after variable intervals. Similarly, musician’s dystonia is described in the literature as a task-specific movement disorder that manifests itself as a loss of voluntary motor control in extensively trained movements while a musician is playing the instrument. For instruments where workload differs across hands, the dystonia is noted in the more intensely used hand. This is considered highly disabling and often results in a terminated musical career. Reporting that this affects 1% of all professional musicians, behavioral factors are cited as extrinsic triggers, specifically the use of maximal fine-motor skills with associated “perfectionist tendencies.”\(^10\) In Tanner’s case–control investigation, 83% of the 150 SD patients studied reported a history of intense occupational voice use during their lifetimes. They pointed out that intense voice use might be analogous to other repetitive fine motor tasks of the hands or limbs that have been shown to contribute to focal dystonias, such as musician’s dystonia.\(^8\)

The prevalence of extrinsic factors identified by patients with rapid-onset SD in our study suggests that some form of external trigger may provoke the development of symptoms in a subset of patients. Thus, SD could be the result of a “two-hit” model of pathogenesis, including both an intrinsic and extrinsic factor.

Regarding specific factors identified in our study, “stress” is liable to highly individual definitions, is relatively common, and has poorly understood neurologic correlates, so it remains extremely difficult to assess its relevance. Nevertheless, to the extent that stress may be understood as a psychological factor, there may yet prove to be a psychologic dimension to SD. The presence of an upper respiratory infection (URI), although common, raises the possibility of a peripheral mechanism in SD because of the strong association of a URI and peripheral cranial neuropathy. The typical dystonic feature of a “geste antagoniste” or sensory trick, and clinical response to botulinum toxin in excess of that expected from a simple end-organ effect, as well as positron emissions tomography (PET) scan evidence of indirect central nervous system (CNS) changes after botulinum toxin injections speaks to the importance of efferent nerves in dystonia. The endotracheal intubation may constitute an end-organ-specific insult analogous to the peripheral trauma identified by Kumar and Jog. On the other hand, the relation of pregnancy and parturition to SD remains obscure but intriguing given its prominence.

Retrospective studies like those that have been done to date are most useful in raising questions and helping formulate hypotheses. However, observations from retrospective investigations are subject to recall bias. Recall bias plays a significant role with regard to recalling both duration of onset and perceptions regarding potential triggers or associations. Especially when considering those patients who described the onset of their disorder as gradual, there is great variation noted in the time course, with a range of symptom development from just more than 1 week’s time to a more prolonged period of over 13.5 years. Unfortunately, our case history format did not require patients to specify the exact stressor when they attributed the onset to “stress”. On the other hand, a number of patients described specific events that were temporally related to onset, such as pregnancy and parturition, intubation, smoking cessation, and initiation of immunotherapy. Such precisely dateable events are less subject to patient impressions.

Prospective controlled studies are necessary to obtain and confirm the data in the present study. Nonetheless, the current sample of 350 subjects reveals highly variable circumstances surrounding the onset of spasmodic dysphonia; this in turn suggests that the pathophysiology of spasmodic dysphonia may be less homogeneous than is generally thought. Future studies that not only clarify exactly the type of stressors identified as triggers, but also specifically quantify the onset and associated events surrounding the onset will help to further our understanding of this chronic voice disorder.

CONCLUSIONS

A subgroup of patients perceive SD to be sudden in onset and associated with specific triggers. This highlights the possibility that certain environmental factors may represent a key component in the pathophysiology of this disorder. These may include peripheral postviral neuropathy, psychological stressors, endotracheal intubation, or end-organ injury. Pregnancy and parturition are identified as a discrete factor that has previously not

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been described. Further study may better define these and other triggers.

BIBLIOGRAPHY