Spasmodic dysphonia

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Introduction

This article includes discussion of spasmodic dysphonia, abductor dysphonia, adductor dysphonia, and spastic dystonia. The foregoing terms may include synonyms, similar disorders, variations in usage, and abbreviations.

Overview

Spasmodic dysphonia is a focal laryngeal dystonia. The more common adductor type typically results in strained effortful speech with breaks in phonation. Abductor spasmodic dysphonia generally causes breathy speech with voiceless pauses. The task-specific nature of this condition means that it may normalize with changes in pitch or volume or with other activities, such as laughing or yawning. Several causative genes have been identified in spasmodic dysphonia. In this article, the author discusses risk factors, clinical features, pathophysiology, treatment, and updates on genetic causes.

Key points

- The diagnosis of spasmodic dysphonia is made clinically based on perceptual voice evaluation.
- Botulinum toxin injections have become the mainstay of treatment for spasmodic dysphonia.
- Advances in genetic studies have allowed causative genes to be identified in some individuals.

Historical note and terminology

Spasmodic dysphonia is a focal dystonia resulting in task-specific, action-induced spasm of the vocal cords. Historically, Tiberius Claudius Drusus Nero Germanicus, who became emperor of Rome 41 AD, has been suspected to have spasmodic dysphonia (Rice 2000). It was first described by Traube in 1871 as a “nervous hoarseness” in a young girl and assigned the label of spastic dysphonia (Traube 1871). The patient only spoke with great effort and “the laryngoscopic examination revealed spastic closure of the vocal cord, whereby the left arytenoid cartilage shifted in front of the right one while probably also the vocal cords were particularly overlapping of each other” (Schaefer and Freeman 1987). Schnitzler may be the first one to suspect organic etiology, in 1895, in 2 patients with “cramping of the vocal cord and forced voice” (Schnitzler 1895), who also had synkinesis of facial muscles and abnormal movements of the arms and legs (Kiml 1965). Schnitzler termed the condition “aphonia spastica” or spastic dysphonia. Due to the lack of other coexisting neurologic deficit, the disorder continued to be considered psychogenic (Heaver 1959; Schaefer and Freeman 1987; Baizabal-Carvallo and Jankovic 2015). A century later, Aronson pointed out the wax and wane characteristic and propose the term “spasmodic” instead of “spastic,” which implies rigidity (Aronson et al 1968a; Aronson et al 1968b). Credit for reviving interest in spasmodic dysphonia as a medical disorder belongs to Dedo with the proposed recurrent nerve resection, which was a bold decision at the time, when most of his contemporaries still believed in a psychiatric etiology (Dedo 1976).

Clinical manifestations

Presentation and course

The onset of spasmodic dysphonia is insidious in the majority of patients (84%), initially presenting as a nonspecific hoarseness. In a smaller proportion, of patients the onset is sudden (Izdebski et al 1984). Some patients report a prolonged laryngitis prior to the onset of dysphonia. In 1 series, the most common inciting events identified by patients

were stress, upper respiratory infection, and pregnancy and parturition (Childs et al 2011; Ankola et al 2013). Patients who had a sudden onset of symptoms were more likely to identify a specific trigger than patients with gradual onset.

In the early stages, the condition may fluctuate in severity. The difficulty is maximal at the beginning of speech and improves with continuation of speech. Uncommonly, stridor is the sole manifestation of spasmodic dysphonia (Tisch et al 2003).

Later, with progressive worsening, spasm occurs throughout. Slowly the laryngeal spasms become persistent and worsen until phonation becomes impossible (Aronson 1990). Spasmodic dysphonia patients often unconsciously speak in a whisper or compensatorily soften their voices. Aronson reported the median time from the initial onset until spasmodic dysphonia developed into its complete state was 1 year (Aronson 1990). In gradual-onset patients, the symptoms reach a plateau within 6 to 9 months. Patients with fluctuating symptoms reached the plateau later, around 23 months (Izdebski et al 1984). Findings from 1 study suggest that at least a subset of patients experience a more protracted worsening of voice symptoms, with one third of participants reporting progression of 4 years or more after the initial onset of symptoms (Tanner et al 2011a).

Spasmodic dysphonia may be divided into 3 types, depending on the vocal cord muscles involved. These include adductor, abductor, and mixed types. Adductor spasmodic dysphonia is the most common form and accounts for up to 82% of cases (Blitzer 2010). The intermittent glottal closure from hyper-adduction of the vocal cords results in voice breaks during vowels and a strangled, staccato-like speech (Ludlow 2009). Other terms used to describe this voice character include squeezed, hoarse, groaning, and jerky.

Abductor spasmodic dysphonia, on the other hand, presents with excessive breathiness, with voiceless pauses and poor generation of volume. There may be prolonged voiceless consonants before initiation of vowels. This type of spasmodic dysphonia occurs in 17% of cases (Blitzer and Brin 1991). The mixed type contains elements of both adductor and abductor spasmodic dysphonia; however, 1 type often predominates.

Spasmodic dysphonia most commonly affects normal speech, typically in the normal talking range of pitch, volume, and speed. Laughing, screaming, and yawning are normal. The fluency may be improved by speaking in a higher pitch, and this is likely related to the task-specific nature of dystonia. Similarly, some patients report that sensory tricks such as touching the larynx, supporting the head, or lying down may improve speech. Voice improvements also occur when the patient is taken by surprise or in sudden danger. Vacation often brings improvement in voice quality. Sometimes improvement is noted with sedative and alcohol use. Patients often report worsening of the voice under stress or on the telephone. The more they concentrate to speak the worse their voice becomes. The voice is vulnerable to anxiety, nervousness, and depression. Physical labor may worsen the voice. Table 1 lists the factors influencing the severity of spasmodic dysphonia (Tisch et al 2003).

Table 1. Factors Influencing Subjective Severity of Spasmodic Dysphonia

	Relieving factor	Aggravating factor
Stress	—	47.3%
Talking on the telephone	—	29.6%
Louder speech	23.7%	4.1%
Yawning	19.5%	—
Higher pitch	18.9%	—
Laughter	18.3%	—
Whisper	14.8%	—
Lower pitch	10.6%	—
Alcohol	10.1%	1.2%
Strong statements	8.3%	—
Singing	7.7%	2.4%
Softer speech	7.7%	—
Morning	7.7%	—
Prolonged talking	2.4%	5.3%
Slower speech	2.4%	—
Swallowing	1.8%	—
Inspiratory speech	1.2%	—

(Tisch et al 2003)

Spasmodic dysphonia has a female preponderance (77.6%), with an average age of onset of 51 years (Patel et al 2015). Spasmodic dysphonia is a focal laryngeal dystonia. Patients with laryngeal dystonia may manifest other clinical features in addition to spasmodic dysphonia, including stridor, dystonic cough, dyscoordinate breathing, paroxysmal hiccups, and paroxysmal sneezing (Payne et al 2014). Spasmodic dysphonia may also occur in the setting of segmental or generalized dystonia. In 1 study, patients with cough, dyscoordinate breathing, paroxysmal sneezing, and hiccups were found to have a higher incidence of extra-laryngeal dystonia (Payne et al 2014). However, it may also occur in the setting of segmental or generalized dystonia. The reported prevalence of extralaryngeal dystonia in patients with spasmodic dysphonia varies from 5% to 14 % (Greene et al 1995; Patel et al 2015). Those patients with spreading dystonia had a mean 3.3 body parts involved, mostly involving the lower face, jaw, tongue, or neck. The mean time before spreading was 7.3 years, with a median time of 4 years, and a range of 1 to 24 years (Greene et al 1995). Tisch and colleagues report the occurrence of extralaryngeal dystonias (including blepharospasm and orofacial and cervical dystonia) preceding the onset of spasmodic dysphonia by a mean of 70.8 months (Tisch et al 2003). In 1 series that included 901 patients with vocal involvement, 82.5% had primary dystonia and 17.5% had secondary dystonia (Blitzer and Brin 1991).

Reports of task-specific laryngeal dystonia occurring only with singing (singer's laryngeal dystonia) have also been described (Blitzer 2010). Respiratory laryngeal dystonia results in adductor spasms during respiration, resulting in inspiratory stridulous noises. They do not usually lead to hypoxia.

Other associated neurologic conditions include dystonic head and neck tremors and postural and action tremors. Comorbid vocal tremor has also been reported (Tisch et al 2003; White et al 2011; Patel et al 2015).

Spasmodic dysphonia may also be associated with increased psychological comorbidity. A study of 44 patients with adductor spasmodic dysphonia revealed substantial degrees of perceived handicap and low perceived control of the condition (Kaptein et al 2010). Rates of anxiety and depression appear to be higher in patients with spasmodic dysphonia and other voice disorders compared to the general population (White et al 2012). Duration of disease was found to be a risk factor for depression in a single-institution case-control study. In a self-administered questionnaire, patients with spasmodic dysphonia reported that their vocal dysfunction negatively affected their work productivity, with the majority reporting some form of presenteeism (Meyer et al 2013).

Prognosis and complications

Spasmodic dysphonia tends to emerge gradually in midlife and then reaches a plateau in terms of severity. Spontaneous remission has not been reported in spasmodic dysphonia as seen with cervical dystonia. Treatment with botulinum toxin injections has altered functional disability but repeated treatment is needed, possibly lifelong.

Clinical vignette

Case 1. A 56 year-old woman complained of difficulty with phonation since age 25. Her condition first started with a cold that never went away. Later, her voice became increasingly hoarse. She was diagnosed with laryngitis and treated with antibiotic but her voice did not recover. Later she was prescribed voice arrest for a month but her condition did not improve. Stress worsened her condition. She took a 1-year leave from her job with the hope that the reduced stress would improve her voice. Speech therapy did not improve her voice. She was hospitalized to observe whether or not she had psychogenic voice disorder. She received psychotherapy without improvement. She returned to work but missed several promotions because of her voice problems. She avoided social events. She no longer wanted to answer the phone and did not want to meet people. She cannot speak comfortably in crowded rooms. She was depressed and became more introverted as she was not able to communicate. Past medical history and review of systems were unremarkable.

Her neurologic examination was normal. Her voice was strained and strangled. She had frequent voice break. There is frequent split of the vowels. When singing the vowel “/i/” from her lowest pitch to her highest pitch the spasm is worse in the midrange. She can scream, laugh, and yawn aloud without spasm. She speaks in whisper easily. When speaking in high pitch, like “Mickey Mouse,” there is no spasm. Blocking of the left recurrent nerve with lidocaine 1% briefly improved her conversational speech.

Case 2. A 42-year-old man referred himself for a voice evaluation because of vocal difficulty that was interfering with

his ministry. For several years, he found it more and more difficult to preach effectively. He noticed that his voice was quite clear at the beginning of the day but became tighter in the afternoon until he was unable to speak at the end of the day. A month previous, he was seen by an otolaryngologist who found nothing wrong with the structures of his voice and said the disorder was nonorganic. His voice examination revealed a pattern characterized by a quality that was forceful, strained, and tight, with tremor, inappropriate pitch or pitch breaks, and breathiness. Otolaryngologic evaluation, including videostroboscopy, showed hyperadduction at the glottal and supraglottal level. On laryngeal electromyography, no abnormal spontaneous activities were found although motor unit action potentials were actively and excessively recruited and continued for seconds after cessation of phonation. No other abnormalities were found on neurologic examination.

Biological basis

Etiology and pathogenesis

The etiological and pathophysiological mechanisms underlying spasmodic dysphonia are not known. Spasmodic dysphonia may occur as an isolated dystonia, with Meige syndrome, or as part of generalized dystonia (Marsden and Sheehy 1982; Jankovic and Ford 1983; Hattori et al 2011; Albanese et al 2013; Marras et al 2016).

Several causative genes have been identified in spasmodic dysphonia, often in the setting of a dystonic syndrome.

THAP1: Mutations in the DYT6 gene *THAP1* typically present with dystonia affecting the cervical, cranial, and upper limb musculature, often with laryngeal involvement (Blanchard et al 2011; Xiromerisiou et al 2012). *THAP1* mutations have also been identified in patients with early-onset generalized dystonia with spasmodic dysphonia (Djarmati et al 2009).

TUBB4: Mutations in the *TUBB4* gene on chromosome 19p13.3-p13.2, which encodes for a neuronally expressed tubulin, cause DYT4 dystonia, originally described in an Australian family (Lohmann et al 2013). This form of dystonia is characterized by prominent spasmodic ("whispering") dysphonia associated with craniocervical dystonia and a "hobby horse" type gait.

ANO3: Mutations in *ANO3* (DYT24) typically present with cranio-cervical dystonia, including spasmodic dysphonia. Mild upper limb dystonia may also be present (Stamelou et al 2014). Tremor is a characteristic feature of *ANO3* mutations, differentiating it from the typical DYT6 phenotype.

GNAL: Mutations in *GNAL* (DYT25) are another cause of cervical dystonia and may be associated with head tremor and spasmodic dysphonia (Balint and Bhatia 2015). Generalized dystonia occurs in approximately 10% of cases. Isolated laryngeal dystonia/spasmodic dysphonia has also been described (Putzel et al 2016).

KMT2B: This syndrome, which is caused by mutations in the *KMT2B* gene, results in a progressive childhood-onset dystonia, with prominent cervical, cranial, and laryngeal dystonia (Meyer et al 2017). It is associated with typical facial features of an elongated face and bulbous nasal tip. MR imaging reveals characteristic findings of subtle symmetrical globus pallidi hypointensity, with a hypointense lateral streak of bilateral globus pallidus externa.

A form of dysphonia similar to spasmodic dysphonia has also been described in dominantly inherited ataxia with dentate calcification, currently assigned the name spinocerebellar ataxia type 20 (Knight et al 2004).

Careful clinical characterization of the dystonic syndrome allows accurate phenotype-genotype correlation and may assist in identifying an underlying genetic diagnosis. Genetic screening of patients with spasmodic dysphonia targeted at mutations in *TOR1A*, *THAP1*, and *TUBB4* have a low diagnostic yield (Groen et al 2011; de Gusmã;o et al 2016).

Other reported associations include neuroleptic exposure, either immediately or as part of the tardive syndrome (Warren and Thompson 1998; Armstrong and Randal 2004), mitochondrial disease (Peng et al 2003), valproic acid administration (with improvement after discontinuation) (Oh et al 2004), central pontine myelinosis (Seiser et al 1998), amyotrophic lateral sclerosis (Roth et al 1996), psychogenic dysphonia (Sapir 1995; Baizabal-Carvallo and Jankovic 2015), late onset spasmodic dysphonia with low arylsulphatase A (Martinelli et al 1995), essential tremor (Lou and Jankovic 1991), palatal myoclonus (Doody and Rosenfield 1990), or trauma (Finitzo et al 1987). Factors associated with an increased risk of spasmodic dysphonia, in small or isolated case controlled studies, include a past history of mumps, blepharospasm, tremor, and intense occupational voice use (Tanner et al 2011b), a personal history of cervical

dystonia, sinus and throat illnesses, rubella, dust exposure (Tanner et al 2012), a family history of voice disorders and tremor (Blitzer and Brin 1991), an immediate family history of vocal tremor and meningitis, and an extended family history of head and neck tremor, ocular disease, and meningitis (Tanner et al 2012).

There is evidence to suggest that spasmodic dysphonia is caused by abnormalities of large-scale brain networks, rather than due to pathology limited to the basal ganglia (Fuertinger and Simonyan 2017). Neurophysiological studies reveal that patients with adductor spasmodic dysphonia have a shortened cortical silent period compared with healthy controls. Similar findings are also observed in patients with focal hand dystonia, suggestive of widespread cortical excitability in both of these conditions affecting the motor cortex representing asymptomatic regions of the body (Samargia et al 2014; Samargia et al 2016). Measurements of excitability over the dominant primary motor cortex during “linguistic” and “non-linguistic” tasks following transcranial magnetic stimulation have been shown to differ in patients with spasmodic dysphonia compared with healthy controls, with restoration of these changes following treatment with botulinum toxin (Suppa et al 2015).

The spasmodic dysphonia patient has abnormal blink reflex recovery, pointing to a loss of inhibitory control (Cohen et al 1989). The difference in R2 amplitude attenuation to electrical and mechanical stimulation suggests that the dystonia involves not only the larynx but also other anatomical structures (Cohen et al 1989). Loss of inhibitory control is also seen in many other focal dystonias (Panizza et al 1990) and the underlying defect may be impaired inhibition at the cortical level leading to a decrease in requirement for activation with voluntary motor commands (Chen et al 1997).

Sharbrough and colleagues described abnormal auditory brainstem responses in 7 of 18 patients with spasmodic dysphonia, indicating slower brainstem conduction along the auditory pathway (Sharbrough et al 1978). In another study of 6 patients with adductor spasmodic dysphonia using the auditory brainstem response, 5 of 6 patients had a compromised capacity of the auditory brainstem to conduct impulses (Finitzo-Hieber et al 1982). In a study of 12 spasmodic dysphonia patients using 3 different auditory brainstem response parameters, 75% were abnormal. Three of the 12 had prolonged wave I-V interpeak latency. Seven had pathologic wave V latency shifts at a high stimulus rate (Schaefer et al 1983). These findings were not confirmed by Middleton in a study of 14 spasmodic dysphonia patients with normal hearing (Middleton et al 1997). Postmortem brainstem examination in 2 patients with spasmodic dysphonia revealed several neuropathological changes compared to controls (Simonyan et al 2010). This included small clusters of inflammation in the reticular formation surrounding the solitary tract and spinal trigeminal nuclei and in the pyramids, in addition to neuronal degeneration and depigmentation in the substantia nigra and locus coeruleus.

Using quantitative topographic electrophysiologic mapping and SPECT, Devous and colleagues suspected that dysfunction of cortical perfusion, cortical electrophysiology, or both, occurred in spasmodic dysphonia (Devous et al 1990). Studying a 59-year-old male patient with adductor type spasmodic dysphonia during phonation with PET, Hirano and colleagues noted remarkable activities during phonation in the left motor cortex, Broca area, cerebellum, and the auditory cortices, whereas the supplementary motor area was not activated (Hirano et al 2001). In normal subjects, significant activities were observed during vocalization in the motor area, Broca area, the supplementary motor area, and the cerebellum, whereas the auditory association area was not activated, even though the subjects heard their own voice (Hirano et al 1996). Comparing with normal activities there were 2 apparent differences, 1 was the lack of activity in the supplementary motor area and the other was activation in the auditory association area. The auditory association area was not activated during normal vocalization, but came to be activated when the speaker's own voice was distorted (Hirano et al 1996; Hirano et al 1997), as is the case with the spasmodic dysphonia strained voice. As the supplementary motor area is known to function for motor planning, programming (Roland et al 1980) is usually activated in normal phonation and damage of the supplementary motor area causes a severe disturbance of voluntary vocalization. Hirano and colleagues suggested that the functional deficit of the supplementary motor area might be related to spasmodic dysphonia (Hirano et al 2001). In a PET study using [11C] raclopride (RAC) to assess striatal dopaminergic neurotransmission, patients with spasmodic dysphonia demonstrated decreased RAC displacement during symptomatic speech production compared with controls, indicating decreased dopaminergic transmission (Simonyan et al 2013). RAC displacement was increased during the unaffected task of asymptomatic tapping, possibly representing a compensatory adaptation of the nigrostriatal dopaminergic system.

When fMRI measurements were performed during vocal motor tasks in patients with laryngeal dystonia and compared with healthy volunteers, reduced activation of primary sensorimotor as well as premotor and sensory association cortices during vocalization in patients with laryngeal dystonia were noted (Haslinger et al 2005). Gray matter volume,

cortical thickness, and brain activation on fMRI were increased in the laryngeal sensorimotor cortex, inferior frontal gyrus, superior or middle temporal and supramarginal gyri, and cerebellum in a study of 40 spasmodic dysphonia patients compared with controls (Simonyan and Ludlow 2012). Phenotype-specific abnormalities have been reported in adductor and abductor forms of spasmodic dysphonia on high resolution MRI and diffusion-weighted imaging. Differences in cortical thickness and white matter fractional anisotropy were observed in the left sensorimotor cortex and angular gyrus and the white matter bundle of the right superior corona radiate (Bianchi et al 2017). In addition, genotype-specific abnormalities were seen between sporadic and familial cases in the left superior temporal gyrus, the supplementary motor area, and the arcuate portion of the left superior longitudinal fasciculus.

Lenticular nucleus hyperechogenicity has been observed on transcranial sonography in patients with spasmodic dysphonia (Walter et al 2014). A study examining a combination of independent component analysis and linear discriminant analysis of resting-state functional magnetic resonance imaging data found abnormal functional connectivity within sensorimotor and frontoparietal networks in patients with spasmodic dysphonia compared with control subjects (Battistella et al 2016). In addition, differences in abnormal functional connectivity appeared to distinguish between the different clinical phenotypes of spasmodic dysphonia, as well as between the genetic and sporadic forms. Differences in cortical surface area have also been described in subjects with spasmodic dysphonia compared with control subjects in regions associated with sensorimotor integration, motor preparation, and motor execution, as well as in areas of processing of auditory and visual information (Kostic et al 2016).

Histologic analysis of the recurrent laryngeal nerves in 2 patients revealed no apparent signs of either destruction or degeneration. The percentage of thin nerve fibers, the diameter of which may range from 5 to 10 micron, however, was higher than in normal controls (Kosaki et al 1999). In another study, recurrent laryngeal nerve removed from patients with spasmodic dysphonia at the time of surgery, using light and electron microscopy, were compared with control recurrent laryngeal nerves (Carlsoo et al 1987). Slight morphometric differences were found between the 2 groups, but these cannot explain causation of spasmodic dysphonia (Carlsoo et al 1987).

Epidemiology"

The prevalence of primary laryngeal dystonia is estimated to be 5.9 per 100,000 (Asgeirsson et al 2006). A study in Ireland found a prevalence of 17.8 per 100,000 for adult onset idiopathic isolated focal dystonia, with 3% of those subjects manifesting spasmodic dysphonia (Williams et al 2017). Spasmodic dysphonia occurs more often in women than in men (Blitzer and Brin 1991; Soland et al 1996; Adler et al 1997). In 1 series, women made up to 79.3% of the population with spasmodic dysphonia (Adler et al 1997). The overall ratio of females to males was 3.8:1. In other reports the female-to-male ratios range from 1.4:1 to 2.6:1 (Blitzer and Brin 1991; Soland et al 1996). Broken down into subgroups, the female-to-male ratio was 4.1:1 for the adductors and 2.2:1 for the abductors. The age at onset was from 13 to 71 years with a mean age of 45 years in 1 series and 39 years in another (Schweinfurth et al 2002). Izdebski and colleagues report in a series of 200 patients with spasmodic dysphonia, the age of onset at 41 years (+13.25 SD) with a range of 6 to 65 years for males. For females, the mean age at onset was similar, at 45.4 years (+13.3 SD), with a range of 7 to 78 years (Izdebski et al 1984).

Prevention

There are no known methods to prevent spasmodic dysphonia.

Differential diagnosis

The differential diagnosis of spasmodic dysphonia is broad and includes both organic and functional disorders. Most of these conditions can be excluded by careful clinical examination; however, in some cases the diagnosis may be challenging. Essential voice tremor and muscle tension dysphonia can cause voice breaks and can form the most important entities of the differential diagnosis. The movement disorder in essential tremor is rhythmic rather than spasmodic, and it often involves pharyngeal and strap muscles. Lundy and colleagues, by applying acoustic and motor speech parameters to the problem, found that unlike spasmodic dysphonia, tremor is more often marked by fluctuations in frequency rather than just in intensity (Lundy et al 2004). Finally, it should be noted that tremor may coexist with dystonia in as many as one third of patients (Koller et al 1994).

Muscle tension voice disorder may also mimic the strained voice quality of abductor spasmodic dysphonia (Houtz et al 2010). Evidence of task-dependent sign expression and intraword phonatory breaks should raise suspicion of adductor

spasmodic over muscle tension dysphonia. The hyperadduction of muscle tension dysphonia is generally sustained and is unlikely to be spasmodic. Neither tremor nor muscle tension dysphonia demonstrate task specificity, although the voice may deteriorate with stress in both conditions. Although differences between muscle tension dysphonia and adductor spasmodic dysphonia have been described on fiberoptic laryngoscopy, phonatory airflow measurement and acoustic analysis, there is currently no single diagnostic test to differentiate these 2 disorders (Roy 2010). The diagnosis is made clinically based on perceptual voice evaluation. In psychogenic dysphonia, there are often a number of atypical characteristics, including loss of normal shouting, yawning, and laughing. The spasms occur also mostly at a certain range when the patient is singing an octave. Psychogenic dysphonia also does not present with tremors (Leonard and Kendall 1999). Spasmodic dysphonia symptoms also occur frequently in association with specific phonetic and phonatory variables, and sometimes in association with sound prolongations and voice arrests. In psychogenic dysphonia, the symptoms are intermittent only in patients with "mild" dysphonias and are more often invariant across phonetic and most phonatory variables and not associated with sound prolongations or voice arrests (Leonard and Kendall 1999). In 1 study of psychogenic speech and voice disorders, occurring in 16.5% of 182 patients with psychogenic movement disorders, stuttering was the most common speech abnormality (n = 16, 53.3%), followed by speech arrests (n = 4, 13.3%), foreign accent syndrome (n = 2, 6.6%), hypophonia (n = 2, 6.6%), and dysphonia (n = 2, 6.6%) (Baizabal-Carvallo and Jankovic 2015).

Vocal cord polyps can cause hoarseness and are seen most frequently in males and smokers. Vocal cord nodules are commonly seen in singers and are the result of vocal abuse and excessive laryngeal use. The voices have more of a hoarse characteristic than the spasmodic component seen in spasmodic dysphonia. There are also reports of stridor resulting from laryngeal dystonia, and in some cases, this may be misdiagnosed as asthma (Tisch et al 2003). Table 2 lists some differential diagnoses of spasmodic dysphonia.

Table 2. Differential Diagnosis for Spasmodic Dysphonia

- Meige syndrome or part of generalized dystonia (Marsden and Sheehy 1982; Jankovic and Ford 1983)
- Wilson disease
- Huntington chorea
- Parkinson disease
- Essential tremors (Lou and Jankovic 1991)
- Gilles de la Tourette syndrome (Lang and Marsden 1983)
- Spinocerebellar ataxia type 20 (Knight et al 2004)
- Mitochondrial disease (Peng et al 2003)
- Amyotrophic lateral sclerosis (Roth et al 1996)
- Palatal myoclonus (Doody and Rosenfield 1990)
- Drug induced dysphonia (Warren and Thompson 1998)
- Tardive dyskinesia (Armstrong and Randal 2004)
- Recurrent nerve paresis
- Strokes
- Vocal cord nodules or polyps
- Neoplasm
- Inflammation of the vocal cord (gastroesophageal reflux, rheumatoid arthritis)
- Infection of the vocal cord
- Trauma (Finitzo et al 1987)
- Psychogenic dysphonia (Sapir 1995)
- Muscle tension dysphonia
- Asthma

Diagnostic workup

Understanding of the nature of the symptoms, including onset, aggravating factors, alleviating factors, associated symptoms, duration of symptoms, and the character of symptoms is the focus of the history and helps lead to a correct diagnosis.

Table 3. Voice History and Symptoms of Spasmodic Dysphonia

Onset of symptoms

- Sudden
- Progressive

- Aggravating/alleviating factors
  - Speaking
  - Singing
  - Stress
  - Prolonged phonation
- Associated symptoms
  - Odynophonia
  - Other neurologic signs
  - Other systemic medical conditions
- Character of symptoms
  - Quality of voice
    - Raspiness
    - Breathiness
    - Strain
  - Flow
    - Decreased breath support
    - Decreased projection
    - Decreased volume
  - Control
    - Loss of pitch control
    - Voice breaks

(Merati et al 2005)

The diagnosis is based primarily on auditory-perceptual features (Ludlow 1995). The best way to assess spasmodic dysphonia is by listening to the patient speak during conversational speech. One should assess the fluidity of the voice, the quality of articulation, and the quality of the voice signal itself. A normal voice should be smooth, without voice breaks or spasm. Listening to the patient reading the "rainbow passage" is also helpful:

> When the sunlight strikes raindrops in the air, they act like a prism and form a rainbow. The rainbow is a division of white light into many beautiful colors. These take the shape of a long round arch, with its path high above, and its two ends apparently beyond the horizon. There is, according to legend, a boiling pot of gold at one end. People look, but no one ever finds it. When a man looks for something beyond his reach, his friends say he is looking for the pot of gold at the end of the rainbow… (Fairbanks 1960; Merati et al 2005).

The "Marvin Williams passage" consists of all voiced sounds; it is another alternative:

> Marvin Williams is only nine. Marvin lives with his mother on Monroe Avenue in Vernon Valley. Marvin loves all movies, even eerie ones with evil villains in them. Whenever a new movie is in the area, Marvin is usually an early arrival. Nearly every evening Marvin is in row one, along the aisle (Merati et al 2005).

Differentiation of adductor or abductor spasms is accomplished by tasks that mix and then isolate the abductor and adductor muscles. Adductor spasm is prominent with voiced sound, such as when a patient performs a long "/i/." Abductor spasms typically will occur during an unvoiced sound that occurs after a voiced sound, such as in the word "sixty-one" (Merati et al 2005).

Many systemic neurologic conditions affect the larynx and often present as dysphonia before manifesting symptoms in other parts of the body. Accurate neurologic diagnosis permits determination of the site of the lesion and can afford early treatment. It is important to differentiate upper motor disease from lower motor disease. A complete and detailed examination is, therefore, a must. Attention should be paid to testing for paralysis but also for fatigability,

agility, coordination, and flexibility during the neurologic examination. Isolation of the abductor muscles is best achieved by having the patient sniff repetitively. Evaluation of the adductor muscles can be performed by having the patient alternate between a sniff and the sound "/i/" repeatedly. The tensor muscles of the larynx are evaluated by having the patient perform the glissando maneuver; that is, having the patient slide from his or her lowest pitch to his or her highest pitch while phonating the sound /i/ and then sliding down from high to low (Merati et al 2005). Agility and fatigability of the vocal folds is assessed by having the patient alternate the sounds /i/ and /hi/ repetitively. Coordination and flexibility are assessed with opposition testing. In this instance, the patient is asked to repeat a phonatory task that involves alternating between adduction and abduction, such as repeating the phrase "/pa/ - /ta/ - /ka/" (Merati et al 2005).

Laryngoscopy is useful in the identification of other vocal cord pathology or the presence of vocal tremor. In one study, laryngoscopic examination did not improve the diagnostic accuracy of spasmodic dysphonia compared with the use of auditory cues alone (Daraei et al 2014).

Examination during speech is most likely to reveal the involuntary laryngeal movements. The larynx is best examined with a flexible nasopharyngoscope as it interferes less with the function of the larynx and permits some limited phonation. Examination with a laryngeal mirror, combined with the necessary traction on the tongue, makes connected speech impossible. It may mask the typical laryngeal features. The larynx should be observed at rest and while performing phonatory maneuvers. Normal vocal folds are predominantly in an abducted position at rest, adducting slightly with expiration, and abducting during inspiration. A median position of 1 or both vocal folds usually implies vocal fold fixation either from paralysis, scar, or joint immobility. A paramedian position is usually associated with vocal fold paresis, scar, or joint hypomobility (Merati et al 2005). Paradoxical vocal fold movement, characterized by adduction during inspiration and abduction during expiration, may be associated with reflux-induced or asthma-induced laryngospasm, anxiety, or tardive dyskinesia. Spontaneous rhythmic activities can be seen with tremor, spasmodic dysphonia, or myoclonic activity. Muscle tension dysphonia (also commonly referred to as supraglottic hyperfunction) occurs when the supraglottic, pharyngeal, and strap muscles are overly recruited during phonation. On examination, this has the appearance of an anterior to posterior or lateral to medial squeezing of the false vocal folds and the pharyngeal muscles. Typically, this behavior is present consistently throughout an entire sample of running speech and singing. When the false vocal folds participate in phonation, it is referred to as dysphonia plica ventricularis. Spasmodic dysphonia produces spasmodic activity of the involved muscles. In adductor dystonia, the spasms result in bursts of glottic compression during phonation. With abductor dystonias, spasmodic abduction of the vocal folds occurs during running speech, resulting in "breathy" voice breaks.

Dedo performed lidocaine blockade of the recurrent laryngeal nerve to determine the potential effects of nerve sectioning (Dedo 1976). Three to 5 mL of lidocaine 1% is injected into the tracheoesophageal groove. A successful blockade results in note improvement in phonation with decrease in spasm for a very short period (a few minutes). The blockade and the positive result can be used to confirm the diagnosis of spasmodic dysphonia.

Stroboscopy is another technique that allows assessment of the vibratory function and architecture of the vocal folds. Small vocal fold cysts and polyps may result in alteration of the voice and stroboscopic examination may help to determine the contribution of the lesion to the patient's vocal complaints. The stroboscopic examination, however, is a complementary test to help guide the evaluation and management of voice disorders. The diagnosis of spasmodic dysphonia can be achieved without it.

Other modalities have also been studied in an attempt to provide a more objective measure in differentiating adductor spasmodic dysphonia from muscle tension dysphonia. The long-term average spectrum (LTAS) assesses the average amplitude spectrum across a selective frequency range and provides information on the spectral distribution of the speech signal over a period of time (Houtz et al 2010). One study suggested that the long-term average spectrum may identify spectral noise differences between muscle tension dysphonia and adductor spasmodic dysphonia in women. The use of neural network and support vector machine-based methods, in combination with a pattern recognition algorithm, has also been studied (Schlotthauer et al 2010). Fine kinemetric analysis from high-speed digital imaging may assist in the clinical differentiation of adductor spasmodic dysphonia and muscle tension dysphonia, although further studies are required (Patel et al 2011).

Other measures of voice quality, which may provide further characterization of spasmodic dysphonia, include the IINFVo (Impression, Intelligibility, Noise, Fluency, and Voicing) perceptual rating scale and the AMPEX (Auditory Model Based Pitch Extractor) acoustical analysis (Siemons-Luhring et al 2009). Measurements in perceptual, acoustic, and

self-assessment dimensions all demonstrate significant improvement in symptoms following botulinum toxin therapy. However, these 3 parameters were found to have poor intrinsic correlation; thus, a tridimensional approach may be preferable (Dejonckere et al 2012). Another study found that the latency between the initiation of thyroarytenoid electrical activity and the onset of phonation was significantly related to the severity of adductor spasmodic dysphonia (De Biase et al 2010). The technique of airflow interruption may also provide additional quantitative information regarding laryngeal function in spasmodic dysphonia (Hoffman et al 2009). One study examining the perceptual structure of adductor spasmodic dysphonia and the acoustic correlates of underlying perceptual factors identified a 2 factor model of adductor spasmodic dysphonia, characterized by hyperadduction and hypoadduction (Cannito et al 2012). Perceived ratings of roughness appeared to be most representative of the hyperadduction factor, whereas breathiness was most strongly associated with the hypoadduction factor. Onset of relative fundamental frequency measures were found to be related to perceived vocal effort in patients with adductor spasmodic dysphonia in another study (Eadie and Stepp 2013).

In the setting of associated clinical features, such as segmental dystonia, tremor, or gait disturbance, careful clinical characterization of the phenotype may provide glues to an underlying genetic dystonic syndrome (see etiology and pathogenesis).

Management

Botulinum toxin. Botulinum toxin injections are the mainstay of treatment for spasmodic dysphonia. The American Academy of Otolaryngology-Head and Neck Surgery as well as the American Academy of Neurology endorses botulinum toxin as primary therapy for spasmodic dysphonia. Blitzer and colleagues performed the first botulinum toxin treatment on a spasmodic dysphonia patient (Blitzer et al 1986; Blitzer et al 1988), and their work was confirmed by a subsequent double-blind study (Troung et al 1991). Meta-analysis of 30, mostly single-blind studies, indicated moderate overall improvement as a result of botulinum toxin treatments (Boutsen et al 2002). The Cochrane Collaboration Review noted that although only 1 study (Troung et al 1991) was double-blind and fulfilled their inclusion criteria out of the nearly 77 reports, most of the studies had similarly positive effects related to the length of treatment, degree of improvement, patient satisfaction, and observed side effects (Watts et al 2004).

Treatment of adductor spasmodic dysphonia results in an average onset of action of 2.4 days. The average peak benefit is 9 days, and this lasts for approximately 15.1 weeks (Blitzer et al 2015). Patients report an improvement to 91.2% of normal (Blitzer 2010). Treatment with botulinum toxin leads to a reduction in spasmodic contractions observed on video laryngoscopy and reduces the voice handicap index score (Esposito et al 2015). Fine wire electromyography has revealed that both the thyroarytenoid and the lateral cricoarytenoid muscles might be affected in adductor spasmodic dysphonia, even if the thyroarytenoid is more predominant (Klotz et al 2004). In contrast, the thyroarytenoid and lateral cricoarytenoid muscles are equally involved in tremor spasmodic dysphonia.

Different protocols have been proposed for injecting botulinum toxin into the thyroarytenoid muscle, either unilaterally or bilaterally. Initial dosages of 1.25 units bilaterally of onabotulinum toxin A resulted in a statistically significant shorter duration of breathiness without significant differences in clinical effectiveness or voice outcome in 1 study, compared with an initial dosage of 2.5 units bilaterally (Rosow et al 2013). Unilateral injection may result in fewer adverse events such as breathiness or hoarseness (Koriwchak et al 1996; Bielamowicz et al 2002). Unilateral injections improve the performance ratio of strong voice interval divided by breathy voice interval (Koriwchak et al 1996). Some studies have shown that the duration of benefit in men is significantly longer and with less swallowing difficulty after bilateral injections (Maloney and Morrison 1994). Upile and colleagues found that low-dose unilateral injections resulted in no significant difference in patient outcome in terms of voice duration, voice score, and complication rate when compared with bilateral injections (Upile et al 2009). The dose of botulinum toxin required for the treatment of adductor spasmodic dysphonia tends to remain stable over time (Rosow et al 2015). There appears to be a lower rate of immunoresistance with botulinum toxin in spasmodic dysphonia compared with other dystonias, and this may be related to the low antigen challenge associated with the low doses used (Mor et al 2016). Higher doses were required for symptom control in female patients with adductor spasmodic dysphonia compared to males in 1 retrospective chart review, although the reasons for this observation are unclear (Lerner et al 2017).

There are different techniques of botulinum toxin injection, including the percutaneous approach (Miller et al 1987), the transoral approach (Ford et al 1990), the transnasal approach (Rhew et al 1994), and point-touch injections (Morzaria and Damrose 2011). In the percutaneous approach, the toxin is given through a Teflon-coated needle connected to an EMG machine. The needle is inserted through the space between the cricoid and thyroid cartilages,

pointing toward the thyroarytenoid muscle. The localization of the needle is verified by high-frequency muscle discharges on the EMG when the patient performs a long “/i/” (Miller et al 1987; Brin et al 1989). Others use laryngoscopic control in addition (Troung et al 1991). One hour to 3 days after the injection, the clinical effects set in, and the patient may experience breathiness for up to 2 weeks, followed by a strong voice. After an effective period of a few months, the spasmodic symptoms slowly return as the clinical effect of botulinum toxin wears off. The duration of effect is dose-related. Although transnasal and transoral techniques may demonstrate lower failure rates than percutaneous EMG-guided methods, they are not as well tolerated (Ludlow et al 1988; Rhew et al 1994; Garcia Ruiz et al 1998; Boutsen et al 2002) and are associated with unacceptable excessive waste of toxin due to the length of the dead volume from the syringe to the needle in the transnasal or transoral techniques. Overall, the advantages and disadvantages of the different techniques are not well defined, and the physician should use the method with which he or she is most comfortable and can most safely perform.

Patients with adductor spasmodic dysphonia and associated vocal tremor may benefit from combined interarytenoid and thyroarytenoid muscle botulinum toxin injections (Kendall and Leonard 2011).

Possible complications from botulinum toxin injections into the thyroarytenoid musculature include breathlessness, throat pain, and dysphagia. Initial functional decline following botulinum toxin injection was observed in 28.5% of cases (Novakovic et al 2011). In a series of 1300 patients over 24 years of age, adductor injections were associated with a 25% risk of mild, transiently breathy voice and a 10% risk of transient coughing with drinking fluids. Local pain, bruising, and itch were reported in less than 1% of individuals (Blitzer 2010). Rarely, bilateral abductor paralysis has been reported and is likely a result of toxin diffusion around the muscular process of the arytenoid to the posterior cricoarytenoid muscles (Venkatesan et al 2010).

Treatment of abductor spasmodic dysphonia is less satisfactory, with patients reporting an improvement to 70.3% of normal (Blitzer 2010). The average onset of effect is 4.1 days, with a peak effect at 10 days. The mean duration of benefit is 10.5 weeks (Blitzer et al 2015). Successful outcomes have been reported using an initial unilateral posterior cricoarytenoid muscle injection, followed by a contralateral injection after 2 weeks if necessary (Blitzer 2010). Simultaneous bilateral posterior cricoarytenoid injections have also shown success in improving vocal outcome measures (Klein et al 2008). The needle is inserted from behind the posterior edge of the thyroid lamina when it is rotated forward and directed toward the posterior cricoarytenoid muscle. The position of the needle can be verified by maximal muscle discharge when patients perform a sniff (Blitzer et al 1992). In another approach, the EMG needle is placed supracricoid under flexible transnasal laryngoscopic surveillance (Rontal et al 1991). The needle is directed along the superior border of the posterior cricoid lamina and between the arytenoid cartilages. For anatomic reasons, the toxin is injected at the area above, to diffuse down into the muscle for therapeutic effects. A refined technique with the needle penetrating through the posterior cricoid lamina into the posterior cricoarytenoid muscle seems to be simpler and has the advantage of direct injection into the muscle (Meleca et al 1997). Adverse effects included exertional wheezing, stridor, dyspnea, and dysphagia.

Botulinum toxin doses. Five different botulinum toxin products are available on the market. Four of these toxins are botulinum toxin type A and include Botox® (Allergan, Inc., Irvine, California, United States), Dysport® (Ipsen Ltd., Slough, Berkshire, United Kingdom), Xeomin® (Merz Pharmaceuticals, United States), and CBTXA (China). Myobloc®/Neurobloc® (Solstice Neurosciences, Inc., South San Francisco, California, United States) is botulinum toxin type B. In the United States, the available preparations include Botox® and Myobloc®/NeuroBloc®. Xeomin® (incobotulinumtoxinA) has been approved for cervical dystonia and blepharospasm. Doses of botulinum toxin used for the treatment of spasmodic dysphonia vary depending on the particular brand of toxin used. In the early literature, the doses of botulinum toxin (Botox®) used for adductor spasmodic dysphonia ranged from 3.75 to 7.5 U for bilateral injections (Brin et al 1988; Brin et al 1989; Brin et al 1998; Troung et al 1991) to 15 U for unilateral injections (Miller et al 1987; Ludlow et al 1988). Up to 50 U per vocal cord has been used (Jankovic et al 1990). Later literature and common practice have recommended the use of lower doses (Blitzer and Sulica 2001). The optimal dose of botulinum toxin varies between patients, and does not appear to correlate with severity of spasmodic dysphonia, age, or gender. In 1 study, BMI and overall health were correlated with a higher effective dose (Young and Halstead 2014). In 1 retrospective study, patients with adductor spasmodic dysphonia who received long-term injections appeared to have an initial reduction followed by stabilization in their dosage (Namin et al 2017). Those who have a fluctuating dosing trend tended to have a shorter interval between injections.

Doses of 1.25 to 1.75 U of Botox® were used in 1 muscle and 0.9 U on the opposite side (Meleca et al 1997). Between

2 and 4 U of Botox® or 12 U of Dysport® on 1 side, and 1 U of Botox® or 3 U of Dysport® on the opposite side are used (Truong and Bhidayasiri 2006). If a higher dose is required for each side, delaying the injection of the opposite side for 2 weeks to avoid compromising the airway is appropriate. One study reported success with unilateral injections of Dysport®, with mean doses of 3.6 ± 0.02 units (approximately 1.2 Botox® units), providing no significant difference in duration of action, voice score, and complication rate compared with bilateral injections using a mean total dose of 6.6 ± 0.02 units of Dysport® (Upile et al 2009). There is very limited experience with botulinum toxin type B (Myobloc®/NeuroBloc®) for abductor spasmodic dysphonia.

Surgery. After recognition of the organic etiology of spasmodic dysphonia, the development of recurrent nerve resection has provided initial remarkable results. Dedo reported the results of sectioning of the recurrent laryngeal nerve in 34 patients (Dedo 1976). The first patient was a woman with a 17-year history of spasmodic dysphonia who had been treated by more than 29 physicians from different specialties without improvement. Blocking of the recurrent nerve with lidocaine improved her spasm. After 5 repeated reproducible results with lidocaine, the patient was operated on with sectioning of the right recurrent laryngeal nerve. Postoperatively, the patient developed essentially normal voice within a week, with the help of speech therapy. Subsequently, 33 additional patients underwent similar procedures from a group of 41 patients who responded to recurrent nerve block. Dedo also performed placebo injections, which did not improve the patient's voice (Dedo 1976). Lidocaine paralysis of the recurrent nerve is a useful indicator for patient selection for recurrent nerve resection (Izdebski et al 1981). Short- and mid-term results were good, with a recurrent rate of 10% to 15%, respectively (Izdebski 1981; Dedo and Izdebski 1983a; Dedo and Izdebski 1983b). Long-term results showed gradual decline in the benefit from the procedure. From the 33 patients followed by the Mayo Clinic for 3 years postoperatively, 36% still had improved voice whereas 64% have “failed voice” (Aronson and De Santo 1983). Of the patients who have failed voice, 48% were worse than before surgery (Aronson and De Santo 1983). Interestingly, 64% of the men were still improved at 3 years compared with only 33% of the women. The failures were attributed to the hyperadduction of the normal true vocal folds and sphincter action of the false vocal folds and sometimes the inferior pharyngeal constrictors. Thinning of the already paralyzed vocal fold using CO2 laser (Dedo and Izdebski 1981), sectioning of the superior laryngeal nerve, and re-resectioning of the recurrent nerve have also been proposed for those with recurrence of symptoms (Dedo and Izdebski 1981; Dedo and Izdebski 1984; Schiratzki and Fritzell 1988; Netterville et al 1991). The mechanism for the recurrence is not yet clearly understood, but an animal study has shown that motor innervation to the thyroarytenoid muscle may also form the external division of the superior laryngeal nerve and is not exclusive by the recurrent laryngeal nerve (ExSLN) (Nasri et al 1997). This additional pathway from the ExSLN to the thyroarytenoid muscle may have important clinical implications in the treatment of adductor spasmodic dysphonia.

Another less invasive procedure involves crushing the recurrent nerve, which appears initially as equally effective. This procedure too suffered from widespread recurrence of symptoms with a success rate of 13% at 3 years (Biller et al 1979).

Selective laryngeal adductor denervation-reinnervation (SLAD-R), first described in 1999 (Berke et al 1999), involves selective denervation of the adductor branch of the recurrent laryngeal nerve. The distal nerve stumps are reinnervated with a nonlaryngeal nerve, generally the sternohyoid branches of the ansa cervicalis nerve (Mendelsohn and Berke 2012). The initial report presented favorable results in 19 of 21 patients. Only 1 patient underwent further treatment with Botox postoperatively (Berke et al 1999). Similar results were also reported in a smaller series in which expert and untrained judges undertook perceptual evaluation of voice quality (Allegretto et al 2003). Additional studies have also reported favorable results after a mean follow-up time of 7.5 years (Mendelsohn and Berke 2012). Functional reinnervation of the vocal cord adductors by ansa cervicalis has been observed 10 years after successful SLAD-R surgery (Deconde et al 2012).

Isshiki and colleagues offered an apparently mechanical solution to the problem of hyperadduction in adductor spasmodic dysphonia with laryngeal framework surgery (Isshiki et al 2000; Isshiki et al 2001). In this procedure, a lateralization laryngoplasty is performed by means of a midline division of the thyroid cartilage. They reported success in a small series with informal judgments regarding voice quality made by both patients and physicians. A further small study using laryngeal framework surgery, or type II thyroplasty, also resulted in significant acoustic and aerodynamic improvement in adductor spasmodic dysphonia (Sanuki et al 2010). Follow-up after 2 to 5 years revealed unsatisfactory outcomes in 7 of 90 cases and were mainly attributed to inadequate technique and in 1 case, an incorrect indication (Sanuki and Isshiki 2009). Surgical mechanical faults appear to be the main cause of unsuccessful outcomes (Isshiki and Sanuki 2010). Another long-term follow-up study (average of 41.3 months) following type II

thyroplasty also demonstrated sustained improvement in voice symptoms and quality of life (Sanuki and Yumoto 2017).

Bilateral thyroarytenoid muscle myectomy is also used in the treatment of adductor spasmodic dysphonia. One study found no difference in the postoperative voice handicap index 10 score between patients who underwent bilateral thyroarytenoid muscle myectomy compared to those who received type II thyroplasty (Nomoto et al 2015). Bilateral thyroarytenoid muscle myectomy appeared to improve strangulation, interruption, and tremor and worsen breathiness when compared to type II thyroplasty. Successful treatment of adductor spasmodic dysphonia has also been reported in a small series of patients using selective lateral laser thyroarytenoid myotomy (Hussain and Shakeel 2010; Gandhi 2014). Sustained improvements in voice handicap index and GRBAS scale have been observed following endoscopic laser thyroarytenoid myoneurectomy (Tsuji et al 2012; Gandhi 2014).

Deep brain stimulation (DBS) for adductor spasmodic dysphonia has been described only in isolated case reports. Deep brain stimulation of the globus pallidus internus (GPi) resulted in improvement of segmental dystonia, including spasmodic dysphonia in 1 patient, with maintenance of symptom control at 10-year follow up (Horisawa et al 2017). Another patient with essential tremor and coincident adductor spasmodic dysphonia treated with unilateral thalamic deep brain stimulation experienced significant improvement in vocal dysfunction (Poologaindran et al 2017).

Other therapeutic modalities. Voice therapy has been reported to be helpful in conjunction with botulinum toxin treatment to retrain a long-term compensatory behavior superimposed on spasmodic dysphonia (Murry and Woodson 1995) although this has not been consistent. Another study did not demonstrate additional benefits in patients receiving voice therapy compared to those receiving botulinum toxin alone, or to those who received botulinum toxin with sham voice therapy. Further studies are required to confirm these results (Silverman et al 2012). Although to a lesser degree than in the past, many spasmodic dysphonia patients are still subjected to psychiatric treatment. Psychotherapy is useful to help coping with social stresses, which can be considerable; the patient, however, needs to be made aware first that his or her condition is an organic disorder and to be treated accordingly either with botulinum toxin or surgery. Acupuncture has not been proven effective by expert raters, although Lee and colleagues found a majority of patients reported subjective improvements in voice production (Lee et al 2003). Electrical stimulation of the thyroarytenoid muscle was reported to improve symptoms in 1 small case series (Pitman 2014). An open-label study of sodium oxybate demonstrated reduced voice symptoms in alcohol-responsive spasmodic dysphonia in those with and without coexistent voice tremor (Rumbach et al 2017).

Special considerations

Pregnancy

Pregnancy has been associated with the sudden onset of adductor spasmodic dysphonia in small studies (Ankola et al 2013).

Anesthesia

It may be difficult to intubate a patient with spasmodic dysphonia. Relaxation and sedation before placement of the endotracheal tube is generally required. Medications, such as dopamine receptor blocking agents, which can worsen dystonia, should be avoided.

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**References especially recommended by the author or editor for general reading.

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ICD and OMIM codes

ICD codes

ICD-9:
Other and unspecified diseases of upper respiratory tract: 478.79

ICD-10:
Other diseases of larynx: J38.7

Profile

Age range of presentation

0-01 month
01-23 months
02-05 years
06-12 years
13-18 years
19-44 years
45-64 years
65+ years

Sex preponderance

female>male, >2:1
female>male, >1:1

Family history

family history may be obtained

Heredity

heredity may be a factor

Population groups selectively affected

none selectively affected

Occupation groups selectively affected

none selectively affected

Differential diagnosis list

Meige syndrome or part of generalized dystonia
Wilson disease
Huntington chorea
Parkinson disease
essential tremors
Gilles de la Tourette syndrome
spinocerebellar ataxia type 20
mitochondrial disease
amyotrophic lateral sclerosis
palatal myoclonus
drug induced dysphonia
tardive dyskinesia
recurrent nerve paresis
strokes
vocal cord nodules or polyps
neoplasm
inflammation of the vocal cord (eg, gastroesophageal reflux, rheumatoid arthritis)
infection of the vocal cord
trauma
psychogenic dysphonia
muscle tension dysphonia
asthma

Other topics to consider

Blepharospasm
Botulinum toxin treatment of neurologic disorders
Childhood movement disorders
Primary dystonia
Psychogenic movement disorders
Rating scales of movement disorders