Spastic dysphonia

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Despite the neurogenic nature of many disorders of phonation, most neurologists are less familiar with these problems than with aphasia and dysarthria. Spastic dysphonia, rarely mentioned in neurological texts and journals, has been known to laryngologists for nearly a hundred years (Traube, 1871), and is the subject of many reports (Arnold, 1959; Bloch, 1965; Kiml, 1965). This paper will describe an auctioneer with spastic dysphonia, and will review the characteristics of this affliction, which is probably neurogenic and related to writer's cramp and other 'occupational neuroses'.

CASE REPORT

A right-handed, white male cattle auctioneer, age 40, was hospitalized because of three years' difficulty in talking. On attempting to speak loudly he had noted hoarseness and weakness of his voice, with tightness of the muscles in his upper abdomen, chest, and throat; increased effort only accentuated the problem. This disturbance, originally present only when auctioneering, eventually affected almost all of his speaking. His speech was normal early in the morning, when extremely intoxicated, and after anaesthesia for laryngoscopy. Numerous vocal cord examinations had disclosed no abnormality. A psychiatrist had found no significant problems and speech therapy had been without benefit. Family and past medical histories contributed nothing.

Examination revealed that the patient was able to cough briskly, and he could phonate normally when producing simple vowel tones or speaking softly. Most of the time, however, despite an appearance of great effort, his voice sounded forced, weak, and low in pitch, with sudden fluctuations in quality, and frequent strangled interruptions. He had similar difficulty in singing or shouting. The abdominal muscles tightened during and even preceding phonation, and while he spoke there was elevation of the larynx and contraction of the chest, neck, and facial muscles. On laryngoscopy, the ventricular bands were seen to participate in phonation.

Routine laboratory studies, radiograph of the chest and electroencephalogram were normal. Pulmonary function studies showed a moderate decrease in expiratory flow rates, perhaps a result of smoking. When his speech was recorded and played back with an 0.25-sec delay as he spoke (delayed auditory feedback) his

language performance improved, a finding considered indicative of a 'functional disorder'. A personality assessment profile yielded a 'psychosomatic pattern'. Consultants in psychology and psychiatry, though unable to identify a conflict area, felt that the speech difficulty was psychogenic.

For several weeks the patient received mild sedation, superficial supportive psychotherapy, speech therapy, breathing exercises, and reading practice with delayed auditory feedback and conditioning shocks to the inappropriately active abdominal muscles. Though objectively unchanged, he claimed to be improving and requested discharge, yet he failed to return for outpatient treatment.

DISCUSSION

There are many causes of abnormal phonation (Wilson, 1940; Morrison, 1955; Arnold and Pinto, 1960; Mayo Clinic and Mayo Foundation for Medical Education and Research, 1963; Adams, 1966; DeJong, 1967), but most can be excluded by a careful history and examination, including laryngoscopy. In our patient, inspection of the larynx showed no structural lesions, such as benign or malignant neoplasm, acute or chronic infection, arthritis, or post-traumatic disorder, and no oedema, ulceration, or granuloma of the vocal cords, such as may result from irritants or habitual voice abuse. Likewise, there were no defects of laryngeal motility to suggest a lesion of the vagus nerve or weakness of laryngeal muscles from strain, laryngitis, or emotional disturbance. The balance of the physical and neurological examination disclosed no indications that the speech disturbance resulted from myxoedema, myasthenia gravis, botulism, tetany, pseudobulbar palsy, senile brain disease, or maladies of the cerebellar or extrapyramidal motor system, such as multiple sclerosis and paralysis agitans.

On the basis of the typical voice changes, inappropriate muscular contractions, negative laryngoscopy at rest, pathological laryngeal contractions on phonation and normal neurological examination, we believe that our patient had a disturbance of

laryngeal function without discernible structural abnormality of the larynx or nervous system. Such a disorder exists, and the vocal pattern is 'so characteristic that the disease could be immediately recognized' (Mackenzie, 1880). The voice is choked, with momentary interruptions of phonation during initial or stressed vowel sounds, and with variations in pitch, intensity, and timbre. Fluctuations exist between normally and abnormally spoken words in the course of a sentence, and the patient often is free of dysphonia in singing or whispering. Great effort is expended in speaking, with displacement of the larynx upward, or less commonly forward and downward, and with habitual tensing of the abdominal muscles preceding and during phonation. At times, synkinetic movements of facial and other somatic muscles are seen as the extreme tension involving the respiratory and laryngeal muscles extends inappropriately throughout the body.

This disorder has been described by many authors, each with his own terminology and interpretation. Spastic dysphonia (Schnitzler, 1875), lalophobia (Coën, 1886), mogiphonia (Fränkel, 1887), aphthongia (Gutzmann, 1898), and psychophonasthenia (Greene, 1938) are only a few of the proposed designations. Arnold (1959) speaks of 'repeated re-discovery... and the invention of new names' and points out that these conditions are indistinguishable. Confusion may have arisen from the over-emphasis of slight variations in symptoms when actually, according to Aronson, Peterson, and Litin (1964), even in a single patient the manifestations may vary considerably from one attack to the next. Although distinctions have been made between hyperfunction and hypofunction of the speech muscles, these authors point out that both conditions may be accompanied by vocal cord nodules, polyps, or contact ulcers, and they indicate that 'hypofunction' may actually result from the fatigue of chronic over-activity and that spastic dysphonia also has a major hyperfunctional component. Like Arnold (1959) they do not distinguish between the many varieties of dysphonia which have been described. For these reasons, it seems preferable to avoid the complex subdivisions of previous reports and to use the term spastic dysphonia in referring to these functional disorders of phonation.

No useful objective methods exist for characterizing abnormalities of phonation. Kiml (1965) has reviewed this topic. A variety of electroacoustic recording techniques has failed to identify any specific voice characteristics. Conventional radiographic examinations of the larynx verify only phenomena which can be observed clinically, and radiocinematography of the diaphragm and larynx has not been done in spastic dysphonia. Laryngo-

scopic findings are shown less frequently in photographs than in drawings which summarize a series of observations. Slow-motion cinematography to show abnormal vocal cord vibrations has not yet been applied to spastic dysphonia and sound movies demonstrating the characteristics of speech disorders are of only documentary value. Measures of respiratory rate, volume, and rhythm in spastic dysphonia show abnormal patterns during speech, but not while breathing quietly or speaking to oneself. These exaggerated and irregular movements fail to correlate with any specific mechanism of disturbed speech. As expected, subglottic pressure is abnormally high while talking. Studies of autonomic function in speech disorders, prompted by the concept of a disturbance in unconscious or automatic control of neural activity, have been naïve in their methodology and of no real value. Laryngeal electromyography in patients with hyperkinetic speech disorders shows an increased number of active motor units. Except for using multi-channel recordings to study the relation of laryngeal and respiratory muscles during speech, or to distinguish neurogenic from functional dysphonias, Kiml foresees little usefulness for this technique.

Laryngoscopy (Morrison, 1955) reveals spasms and irregular contractions of vocal cord tensors with strong adduction of the cords, and opposition of the ventricular bands may be seen as in our patient. The glottis closes or moves irregularly and respiratory muscle activity is similarly ill-controlled. Phonation may be painful, and occasionally the patient cannot make any sound because muscle spasms prevent adduction of the vocal cords, although they may adduct normally in coughing.

Views regarding the aetiology of spastic dysphonia vary. Adams (1966) proposes an unidentified neurological cause, discarding the notion of a psychogenic origin and emphasizing the uselessness of psychotherapy. Robe, Brumlik, and Moore (1960) object to the concept that spastic dysphonia is a single entity, insisting that various organic disorders involving the pathways for speech production are responsible. Though they report 10 patients with abnormal historical, neurological, and E.E.G. findings in support of their contention, the relevancy and interpretation of some of their data seem questionable. Critchley (1939) lists as causes of spastic dysphonia cerebellar, pyramidal, and extrapyramidal disease. On the basis of brain stimulation studies in monkeys (Furstenberg, 1958) which showed that laryngeal movements could be evoked by stimuli outside the primary motor centres, Arnold (1959) views spastic dysphonia as regression to a subcortical mechanism of phonation, and Heaver (1960) talks of the correlation between

psychological and neurophysiological regression, with thalamic emotional influences suppressing the forebrain so that the hindbrain usurps the act of vocal expression. These concepts of 'regression' are confusing and poorly substantiated.

Other authors who consider dysphonia a 'functional' disorder vary in their emphasis. Greene (1957) feels that highly-strung, nervous persons, unable to relax, reflect tensions in a strained voice pattern. Gardner (1958) discusses 'executive's dysphonia', a disorder presumably due to misuse of the voice and responding favourably to treatment. Brodnitz (1963) likewise reports success with a similar group, pointing out that this disturbance occurs most commonly in men holding highly competitive and responsible positions productive of emotional stress. He also describes (Brodnitz, 1962) an acute psychogenic disturbance with abrupt onset of complete aphonia and frequent cure by a single psychotherapeutic effort, and a more chronic disorder yielding slowly to any form of treatment. Such problems, he says, rarely result simply from physical strain on the voice. However, Pradillo (1957) has described painful laryngeal cramps occurring with fatigue in voice professionals who had no other symptoms, and Fränkel (1887) and Gutzmann (1898) noted similar cramping as part of the syndrome of spastic dysphonia. Fränkel felt this was at times an occupational disease and he derived his term for it, 'mogiphonia', from the word 'mographia' used at that time to denote writer's cramp.

More recently, Morrison (1955) and DeJong (1967) have also commented on the similarity between the functional speech disorders and writer's cramp. Wilson (1940) does not mention disorders of speech among the category of craft or occupational cramps, in which he includes writing, sewing, piano and violin playing, and operating a telegrapher's key. Nonetheless, he emphasizes that in most cases of writer's cramp, there is no true cramp but rather a 'conflicting and inappropriate simultaneous activation of many muscles', and this observation seems equally true of spastic dysphonia, in which genuine cramps are rare and there is instead a 'disorder of pneumophonic coordination' (Arnold, 1959). Unfortunately, the literature on writer's cramp and related disorders sheds almost no light on the aetiology of these conditions, which are called 'occupational neuroses' by Wilson (1940) and termed 'functional' in our preceding text. The words 'neurosis' and 'functional' have assumed in recent years a psychiatric meaning, but it should be recalled that in their original context they implied a physiological origin without a demonstrable disturbance of structure. Wilson postulates a cortical disturbance in which the neurones have become

'irritably weak' for certain combinations of movement. Liversedge (1961) contends that such disturbances have a physiological basis, being primarily due to an 'electro-mechanical abnormality of structural neural mechanisms'. He strongly denies the psychological origin of the conditions, quoting Critchley (1954) that 'to look upon the craft palsies as problems in psychopathology has not proved satisfying even to psychiatrists'. His patients demonstrated a full spectrum of mental symptoms, indicating that no single, prime emotional factor could satisfactorily explain the condition.

Treatment in spastic dysphonia has taken many approaches, including classical speech therapy, programs of respiratory and total body exercise, physical therapy, and psychotherapy. There is as much variability in the response to treatment as there is in the reported clinical features. Gardner (1958) and Landes (1962) each report excellent results. Brodnitz (1963) distinguishes between hyperfunctional disorders and spastic dysphonia, finding good results in treating the former group but a poor outcome in the latter. However, it seems probable that these groups are not really qualitatively different and that spastic dysphonia is simply a more severe variety of hyperfunction. Bloch (1965), Heaver (1960), and Kiml (1965) agree on the poor prognosis in spastic dysphonia and stress the tendency of these patients to refuse or interrupt therapy, to do poorly if treated, and to relapse if improvement occurs. Heaver's psychoanalytic view blames the failures on ambivalence of the patient, but, in view of our ignorance in this area, the physician should probably shoulder a generous share of the responsibility when treatment is unsuccessful. The unsatisfactory therapeutic outlook of spastic dysphonia is shared by writer's cramp and the other professional neuroses. The suggestion by Wilson (1940) of a 'long rest' from the movement in question or 'complete abstinence' seems hardly applicable to the act of talking. Liversedge (1961) reports some good results in writer's cramp with conditioned reflex therapy, using pens so constructed that excessive pressure by the thumb and fingers completed an electrical circuit and administered a painful shock to the opposite hand with immediate inhibition of the pressure. A similar form of therapy with shocks applied to respiratory muscles was attempted in our patient, but his termination of treatment made it impossible to evaluate the results. This type of management should be considered for future cases.

SUMMARY

Spastic dysphonia, a striking abnormality of voice production, is described in an auctioneer. Excessive

expiratory pressure against a tightly constricted laryngeal sphincter results in a choked quality of the voice, variable in the course of a sentence, most marked in the stressed vowels, frequently absent in singing or whispering, and often accompanied by evidence of inappropriate and excessive synkinetic activity of facial and other body musculature. There are no laboratory techniques helpful in the evaluation of this problem, and the diagnosis is based strictly on clinical findings. Though often felt to be of emotional origin, the disorder responds poorly to psychotherapy. It is proposed that this condition is related to writer's cramp and similar problems, and that the origin resides in a physiological rather than a psychological disturbance. Though all forms of treatment have had poor results, conditioned reflex therapy merits additional investigation.

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