Journal of Neurology, Neurosurgery, and Psychiatry, 1978, **41**, 361–365

Clinical aspects of spasmodic dysphonia

M. J. AMINOFF, H. H. DEDO, AND K. IZDEBSKI

From the Departments of Neurology and Otolaryngology, School of Medicine, University of California, San Francisco, California, USA

SUMMARY The clinical features of 12 patients with spasmodic dysphonia are described. In 11 patients, the voice was strained, harsh, tight, and tremulous, and was low in volume and pitch. Speech, which was sometimes barely intelligible, was interrupted by irregular stoppages and catches of the voice; it required considerable effort, and was accompanied by facial grimacing. The dysphonia was part of a more widespread neurological disorder (idiopathic torsion dystonia) in one case, while it coexisted with blepharospasm in another, and with postural tremor in two. There was a buccolingual dyskinesia in another of these 11 patients, but this may have been related to her previous drug regime. In the twelfth patient, who had a familial tremor, the voice was characterised by marked breathiness, with intermittent aphonia. The disorder is probably due to a focal dystonia of the laryngeal musculature, and this would be consistent with the type of neurological disorders that were associated with it in our cases. Symptomatic benefit follows the therapeutic division of one of the recurrent laryngeal nerves, in selected cases.

The purpose of this paper is to describe some of the clinical and neurological features of a disorder of phonation which, though uncommon, can be socially crippling to those afflicted with it, and to discuss its nosological status. First described by Traube in 1871, the condition has received scant attention in the neurological literature, perhaps because in the great majority of cases it has erroneously been regarded as psychogenic in nature. Many of the laryngologists to whom affected patients are directed follow Schnitzler (1875) in referring to the disorder as "spastic dysphonia". This term is somewhat misleading, however, since it implies an underlying involvement of corticospinal pathways which is not substantiated on clinical grounds. Recent authors—for example, Aronson *et al.* (1968b)—have, therefore, preferred the designation of spasmodic dysphonia, and although this term is not entirely satisfactory it will be used here in the interests of uniformity.

In the last three years, this hospital has become a major referral centre for patients with spasmodic dysphonia, and a new surgical approach to their treatment has been introduced into practice (Dedo, 1976). For the purpose of this study, 12 consecutive patients were evaluated clinically by a neurologist (MJA), as well as by the laryngologist (HHD) to whom they had been referred. Furthermore, in order that the response of these patients to treatment might be assessed objectively, tape recordings were made of their speech, phonation, and articulation when they presented here, and also after they had been treated by operation.

Address for correspondence and reprint requests: Dr M. J. Aminoff, Department of Neurology, School of Medicine, University of California, San Francisco, California, 94143, USA.

Accepted 21 November 1977

Clinical findings

There were 10 females and two males among the 12 patients studied (Table), and they ranged in age from 31 to 71 years. The duration of their phonatory disturbance varied from four to 23 years. In no instance was there any significant abnormality of the perinatal, developmental, or past history, except as indicated below, nor any history of preceding intake of drugs known to cause a movement disorder.

The phonatory disturbance developed over several months except in cases 2 and 12 in whom it seemed to become established rather abruptly over a few days. After its development, it showed little change or progression with time, although it did show some variation in its severity during the day, being worse when patients were tired or emotionally upset.

Table *Clinical features of the patients studied*

Case number	*Sex*	*Age (yrs)*	*Duration of dysphonia (yrs)*	*Coexisting disorders*
1	F	60	20	Postural tremor
2	F	60	4	Buccolingual dyskinesia
3	F	48	5	
4	F	52	8	Blepharospasm
5	M	31	18	Idiopathic torsion dystonia
6	M	50	5	
7	F	60	9	
8	F	59	9	
9	F	57	8	
10	F	61	2	
11	F	54	19	Postural tremor
12	F	71	23	Postural tremor (familial)

In one patient (case 12) the disturbance was characterised primarily by breathy, quavering phonation and intermittent aphonia. In all our other cases, the voice produced during attempted speech or phonation was "tight", "strained" and "hoarse" in quality, and low in volume and pitch. A superadded tremor (slight in cases 2 and 4) led to a tremulous quavering of the voice. The normal rhythm and tempo of speech were interrupted, and abnormal voice catches and stoppages were frequent except in cases 4 and 8. Most patients preferred to talk in whispers, which they were able to do perfectly easily and well; when obliged to use a talking voice, speech was accompanied by facial grimacing, by tic-like contractions of the neck and shoulder muscles, by obvious distress, and by frequent clearing of the throat in an attempt to obtain a better phonatory sound. Four patients attempted phonation during the inspiratory phase of the respiratory cycle. A mild degree of dysarthria was present in case 5, but not in any other patient.

All patients were able to whisper, phonate in falsetto mode, laugh, cough, or cry normally and without any difficulty.

Full otolaryngological evaluation, including indirect laryngoscopy, revealed no structural abnormality. In case 4 there was a history of blepharospasm which had persisted unchanged since its onset one year before the development of any disturbance of voice. In three cases a history of tremor was obtained. In case 1 this was said to have become troublesome five years after the onset of the phonatory disturbance, and to have been troublesome intermittently ever since, while in case 11 it was first noticed one year after the dysphonia. In both instances examination revealed a fine postural tremor of the outstretched hands similar in character to benign essential tremor. The third patient (case 12) had developed her tremor at least 20 years before her voice became disturbed, and reported that her father and two of her sibs had been similarly affected. Examination revealed a gross postural tremor of both hands as well as titubation.

One patient (case 5), who was not Jewish, had developed dystonic posturing of all his limbs when 13 years old, and soon afterwards involvement of his axial musculature and a disturbance of his voice. He was investigated at a number of different neurological centres where a diagnosis of idiopathic torsion dystonia was made, and eventually, when aged 21 years, underwent bilateral cryothalamectomy which benefited his limb symptoms but did not help his dysphonia.

Detailed neurological assessment revealed no abnormalities in any other patient except one (case 2) who exhibited a buccolingual dyskinesia of uncertain duration but which may have related to the treatment with chlorpromazine and trifluoperazine that she had received elsewhere for her dysphonia.

The electroencephalogram was recorded in all except cases 4 and 12, but in no instance was it clearly abnormal.

Treatment

No attempt was made to treat our patients medically, since drug treatment of this disorder has been consistently unsuccessful over the years, and most patients had already been tried without benefit on a number of different drug regimes before referral here. The emphasis was rather on surgical treatment to improve vocal quality.

Excessive adduction of the vocal folds is thought to occur during speech in patients with spasmodic dysphonia characterised by a tight, strained voice, and Dedo (1976), therefore, tried the effect of deliberately sectioning one of the recurrent laryngeal nerves as a therapeutic manoeuvre in such circumstances. In view of the beneficial effect that followed, the same surgical approach was adopted in the management of most of the present cases.

After initial evaluation, the right vocal fold was temporarily paralysed by infiltrating the recurrent laryngeal nerve at the level of the cricothyroid joint on that side with 1% lidocaine. Paralysis usually lasted between 10 and 30 minutes, and was confirmed by indirect laryngoscopy performed two or three minutes after the injection. If it led to an improvement in the voice, surgical division of one recurrent laryngeal nerve was performed as an elective procedure on a subsequent occasion. The

surgical technique has been described elsewhere (Dedo, 1976).

One of the patients in the present series (case 12) was refused operation because her speech disturbance was characterised mainly by breathy phonation with intermittent aphonia. This kind of disturbance, which has tentatively been attributed (Aronson, 1973) to predominant involvement of the laryngeal abductors, would be exacerbated rather than improved by section of the recurrent laryngeal nerve. In 10 patients, the nerve was sectioned and a segment removed, while in the remaining patient (case 6) it was crushed rather than sectioned, at his own request.

Among the 10 patients in whom the nerve was sectioned, all except one (case 5) showed undoubted improvement in vocal quality as judged subjectively, and by two "blind" independent observers who listened to the tape recordings made of patients' voices before and after treatment. In seven cases, the improvement was dramatic. The tight quality of the voice was lost, clarity of tone was regained, pitch control was improved, and facial grimacing was eliminated. In another two patients (cases 3 and 7) phonation was undoubtedly improved but remained abnormal. In the remaining patient (case 5) there was initially some improvement in vocal quality and a reduction in facial grimacing, but after about two months his speech worsened again. All the patients have now been followed for between one and two years since their operations, and there have been no further developments. The coexisting buccolingual dyskinesia in case 2, blepharospasm in case 4, and postural tremor in cases 1 and 11 were not affected by the operative procedure.

The patient (case 6) in whom the nerve was crushed initially showed marked improvement in the quality of his voice and the ease with which he could use it. About two months later, however, his symptoms reverted to their preoperative severity, and it is, therefore, intended to readmit him for nerve section in the near future.

Histology of the 10 excised nerve specimens revealed no abnormality in any instance.

Discussion

The nature and aetiology of spasmodic dysphonia is not fully understood. Although many still believe that the disorder is psychogenic, in none of our patients was there any overt psychiatric disturbance. Moreover, the failure of their symptoms to remit even temporarily despite changes in personal circumstances or medical treatment is against this view, as is their uniform response to surgical treatment, and the failure of other symptoms to develop after such treatment in place of the phonatory ones.

Critchley (1939) described three patients who spoke with a "constrained, forced, and barely intelligible" voice, in whom he was unable to make a confident diagnosis. One of his patients also had spasmodic torticollis, and another may have had a familial tremor, although it is difficult to be certain about this from the published description. Frequent tic-like movements of the face, neck and jaw—and even of the shoulder girdle and upper arm—were said to occur during speech. All of the 10 patients reported by Robe *et al.* (1960) were said to have some neurological signs, but only in four could a specific diagnosis be made, and insufficient details were provided by the authors to permit independent assessment. Aronson *et al.* (1968a) reported facial and tongue twitches in eight of 27 patients, but it is not clear whether these were tic-like compensatory movements occurring during speech, or whether they occurred independently of speech. Interestingly, six of their patients had a static tremor of the head or hand, and this led them to postulate a relationship with the essential tremor syndrome.

McCall *et al.* (1971), using videofluoroscopy, showed that spastic dysphonia could occur either as a manifestation of an isolated, phonatory-related, tonic spasm of the larynx, or as part of a movement disorder affecting the pharyngeal and laryngeal muscles during quiet respiration as well as during speech. Patients exhibited atypical movement patterns of the pharynx and larynx that were either tremor-related or seemed to be manifestations of dystonia. This accords with the results of subsequent electromyographic studies of the extrinsic laryngeal muscles, since these have revealed the occurrence of avocal contractions of the extrinsic laryngeal muscles in patients with spasmodic dysphonia (Rabuzzi and McCall, 1972).

Our own findings would certainly suggest that spasmodic dysphonia should be regarded as a focal dystonia of the laryngeal musculature. Thus, it may occur in patients with idiopathic torsion dystonia, as in our case 5, or it may be associated with other, related conditions attributed to abnormal function of the extrapyramidal motor system such as blepharospasm (case 4). None of our patients developed orofandibular dystonia, however, in contrast to some of those reported by Marsden (1976). The high incidence of tremor among our patients with spasmodic dysphonia accords with the high incidence reported in patients with torsion dystonia (Johnson *et al.*, 1962; Larsson and Sjögren, 1966; Marsden and

Harrison, 1974), or related movement disorders such as spasmodic torticollis (Couch, 1976).

Little attention has been devoted in the published literature to the pathological basis of idiopathic torsion dystonia or the various associated disorders. It is generally accepted that these conditions are related to dysfunction of the basal ganglia, and we can only assume that this is the case in many patients with spasmodic dysphonia. However, structural abnormalities have been reported in 30% of the recurrent laryngeal nerve specimens obtained during the treatment by nerve section of patients with spasmodic dysphonia (Dedo *et al.*, 1977), although not in any of the cases in the present series, and whether these changes can be related to the dysphonia is uncertain.

Two main types of spasmodic dysphonia were described by Aronson (1973), depending on whether involvement of the adductors or abductors of the vocal folds predominated. In the former, the vocal folds were said to be adducted excessively during phonation, so that the voice sounded strained, while in the latter the vocal folds were abducted excessively, leading to uncontrolled widening of the glottis, and thus to intermittent aphonia and breathy phonation. The disorder of 11 of our patients was of the adductor type, and their voices had a number of features in common. They were strained, harsh, and tight, often quavered tremulously, and were low in pitch and volume. Speech required considerable effort, was frequently unintelligible, was sometimes produced during inspiration, was accompanied by facial grimacing, and was interrupted by irregular stoppages and catches of the voice. In the remaining patient (case 12) the disturbance was very different, the voice being characterised by marked breathiness as in Aronson's (1973) abductor type of spasmodic dysphonia. In no instance was there any disturbance of whispering, laughing, coughing, crying, moaning, or sustained phonation beyond the normal speech range.

In most series the sex incidence is approximately equal, although there was a preponderance of females among our patients. The disorder usually begins in adult life, without obvious cause, and investigations fail to reveal any evidence of an underlying metabolic or structural disturbance of the nervous system.

Indirect laryngoscopy revealed no abnormality in any of our patients, but this procedure usually permits the vocal folds to be visualised only during sustained, highpitched (falsetto) phonation, which patients are anyway able to perform normally.

Spontaneous remission rarely, if ever, occurs, and the results of medical treatment have generally been unsatisfactory. Because a lesion of the recurrent laryngeal nerve leads to retraction from the midline of the vocal fold that it innervates, Dedo (1976) tried the effect of deliberately sectioning this nerve as a therapeutic manoeuvre in patients with spastic dysphonia of the adductor type. The procedure was performed in the belief that excessive adduction of the other vocal cord during speech would carry it across the midline to the deliberately paralysed one; because the mobile vocal fold would be at the limit of its excursion, closure of the folds would not be as tight as previously. It was undertaken only if phonation improved significantly when one vocal fold was temporarily paralysed by topical infiltration of local anaesthetic about the recurrent laryngeal nerve. Clarity of tone, improvement in control of pitch, and elimination of associated movements occurred in all of 34 patients in whom the nerve was sectioned. In no case did speech subsequently revert to its preoperative state, but several patients are reported to have spoken with a mild residual hoarseness, and two were left with rather breathy voices. The potentials, hazards, and complications of the procedure have been discussed by Dedo (1976), and need not be recapitulated.

In the present series, nine of the 10 patients in whom one of the recurrent laryngeal nerves was sectioned undoubtedly benefited as judged both subjectively, and objectively by "blind" observers. This permitted their rapid return to a useful way of life, often after they had been disabled for many years.

References

Aronson, A. E. (1973). *Psychogenic Voice Disorders. An Interdisciplinary Approach to Detection, Diagnosis and Therapy*. W. B. Saunders: Philadelphia.

Aronson, A. E., Brown, J. R., Litin, E. M., and Pearson, J. S. (1968a). Spastic dysphonia. I. Voice, neurologic, and psychiatric aspects. *Journal of Speech and Hearing Disorders*, **33**, 203–218.

Aronson, A. E., Brown, J. R., Litin, E. M., and Pearson, J. S. (1968b). Spastic dysphonia. II. Comparison with essential (voice) tremor and other neurologic and psychologenic dysphonias. *Journal of Speech and Hearing Disorders*, **33**, 219–231.

Couch, J. R. (1976). Dystonia and tremor in spasmodic torticollis. In *Advances in Neurology*, volume 14, pp. 245–258. Edited by R. Eldridge and S. Fahn. Raven Press: New York.

Critchley, M. (1939). Spastic dysphonia ("inspiratory speech"). *Brain*, **62**, 96–103.

Dedo, H. H. (1976). Recurrent laryngeal nerve section for spastic dysphonia. *Annals of Otology, Rhinology and Laryngology*, **85**, 451–459.

Dedo, H. H., Izdebski, K., and Townsend, J. J. (1977). Recurrent laryngeal nerve histopathology in spastic dysphonia: a preliminary study. *Annals of Otology, Rhinology and Laryngology*, **86,** 806–812.

Johnson, W., Schwartz, G., and Barbeau, A. (1962). Studies on dystonia musculorum deformans. *Archives of Neurology (Chicago)*, **7,** 301–313.

Larsson, T., and Sjögren, T. (1966). Dystonia musculorum deformans. A genetic and clinical population study of 121 cases. *Acta Neurologica Scandinavica*, Supplementum 17, 1–232.

McCall, G. N., Skolnik, M. L., and Brewer, D. W. (1971). A preliminary report of some atypical movement patterns in the tongue, palate, hypopharynx, and larynx of patients with spasmodic dysphonia. *Journal of Speech and Hearing Disorders*, **36,** 466–470.

Marsden, C. D. (1976). Dystonia: the spectrum of the disease. In *The Basal Ganglia.* Edited by M. D. Yahr. *Research Publications: Association for Research in Nervous and Mental Disease*, **55,** 351–367. Raven Press: New York.

Marsden, C. D., and Harrison, M. J. G. (1974). Idiopathic torsion dystonia (dystonia musculorum deformans). A review of forty-two patients. *Brain*, **97,** 793–810.

Rabuzzi, D. D., and McCall, G. N. (1972). Spasmodic dysphonia: a clinical perspective. *Transactions of the American Academy of Ophthalmology and Otolaryngology*, **76,** 724–728.

Robe, E., Brumlik, J., and Moore, P. (1960). A study of spastic dysphonia. Neurologic and electroencephalographic abnormalities. *Laryngoscope*, **70,** 219–245.

Schnitzler, J. (1875). Cited by Arnold, G. E. (1959), Spastic dysphonia: 1, changing interpretations of a persistent affliction. *Logos*, **2,** 3–14.

Traube, L. (1871). Cited by Arnold, G. E. (1959), Spastic dysphonia: 1, changing interpretations of a persistent affliction. *Logos*, **2,** 3–14.