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Title: Morell Mackenzie's contribution to the description of spasmodic dysphonia

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Abstract

Objectives: Since the middle of the 20th century most discussions of Spasmodic Dysphonia reference a paper by Ludwig Traube published in 1871 as the first historical citation, crediting him with priority for this clinical syndrome. However, our recent research has determined that the original observation by Traube was published in 1864 and does not in fact describe what is currently recognized as SD. It appears that many clinics throughout Europe and North America were investigating and publishing observations on a range of voice disorders..

Methods: The wider context of work on laryngeal disorders in the 1860s-1870s is considered. One of Traube's contemporaries, Morell Mackenzie made significant contributions to the understanding of laryngeal movement disorder and its consequences for the voice. These will be examined to gain a clearer focus on the characterization of this disorder.

Results: The clinical descriptions published by Morrell Mackenzie in the 1860s provide details which conform quite closely to our current day understanding of SD.

Conclusions: The citation of Traube's "hysterical" patient links to mid-20th century views of the functional nature of SD and the utility of psychiatric treatment. The description presented by Mackenzie is consistent with current views of SD as a movement disorder.

Key words: Dysphonia, Vocal Cord Dysfunction, Spasmodic Dystonia, Voice Disorders, Movement Disorders, 19th century History of Medicine

Introduction

Spasmodic dysphonia (SD), also referred to as Laryngeal Dystonia, is currently described as an idiopathic focal dystonia affecting the intrinsic muscles of the larynx. The National Institute of Health (NIH), USA classifies it as a Rare Disease with an occurrence of approximately 14 cases per 100,000. It is predominately found in women (2.5:1) with 30-50 years as the peak onset age. Vocal symptoms range from occasional difficulty to sustained inability to phonate. There are two phenotypes: a more common adductor type SD and the rarer abductor type, however mixed cases of both types do also occur. Vocal tremor often coexists ¹.

The emblematic case typically referenced as being the first description of SD is credited to the pulmonary specialist Ludwig Traube (1818-1876) in 1871 ². Traube's clinical description characterized it as a "Spastische Form der nervösen Heiserkeit" (a spastic form of nervous hoarseness). The patient was a young girl suffering from Typhus whose voice was described as being very hoarse, and nearly aphonic. She could only intermittently, and with great effort, produce high falsetto vocal utterances. Laryngoscopic examination revealed spasmodic closure of the glottis, with the left arytenoid cartilage moved over the right while the epiglottis was bent steeply to the back. There was extreme muscle tension of the vocal cords, with protrusion of the tongue and lips during speech. Hand tremor was also present. As the patient regained her strength, her hand tremor and spasms of the lips and tongue reduced and her voice returned over a period of a few days. This case appears to have gained priority through the influential writings of ENT clinicians in the mid-20th century. Our historical research, published in an earlier issue of this journal³, has revealed that it was actually recorded several years earlier ⁴.

This case clearly contains feature that do not correspond to our current picture of this vocal disorder. Gilman and Gilman ⁵ point out that when doing historical research "...one must

be careful not to presuppose that we easily could translate the diagnostic categories of the time into our 21st-century diagnostic categories. ‘Updating’ diagnoses based on the scant information in case histories, whereas tempting, presupposes a continuity of diagnosis which does not exist, assumes that medical terminology has been static, and that there is no cultural context for diagnosis”⁵. However, these cautions do not apply to the historical picture presented here. We argue that a competing contemporaneous clinical characterization of SD provided by Morrell Mackenzie (1837-1892) in 1868 does conform to our current clinical picture. While Traube⁶ is cited regularly in the 20th and 21st century literature, Mackenzie’s work appears to have been overlooked. Our objective is to present the work of Mackenzie within the European context of clinical laryngological research in the 1860s to highlight his significant original contribution to SD. We argue that it is Mackenzie and not Traube that should be given priority for the clinical description of this voice disorder.

Historical Context

The timeframe considered here will take as its starting point the mid-19th century. One key factor which contributed to the development of understanding of this clinical entity was the newly devised laryngoscope. This technological innovation was developed in the first half of the 19th century and underwent a series of refinements that provided the opportunity for direct viewing of the larynx and thus more detailed descriptions of voice disorders by physicians in Breslau, Vienna, Berlin and London from the 1860s onwards. The wider historical context of European medicine in the 19th century was influenced by political and economic upheaval, although the local circumstances were quite different in France and Germany. In general, it could be said that medical science was developing in to a more rational, mechanistic and evidence based approach. There was increasing interest in laboratory physiology and the

regularization of clinical examinations informed by a developing patho-anatomical approach ⁷. Before this time, physicians could only surmise the mechanics of the larynx from examination of dissected material from cadavers ⁸.

The history of the development of the laryngoscope which made direct viewing possible in living people for the first time is contested. Although the Spanish singer Manuel Garcia (1805-1906) is often credited with this invention in 1855, others point to the English physician Benjamin Guy Babington, F.R.S (1794-1866) as the creator of the first instrument to view the larynx in 1829 ^{9,10}. In Budapest, the physiologist Johann Czermak's (1828-1873) significant modifications to the laryngoscope that better concentrated the light for viewing and made it easy to use for a wider group of clinicians was also widely acknowledged ¹¹. There was a competing model produced by the Viennese neurologist Ludwig Türck (1810-1868), however perhaps due to his death in 1868, he did not have ongoing impact on the specialty. In addition to these clinical centres in Vienna and Budapest, Ludwig Traube had a clinic in Berlin which also investigated diseases of the throat. These clinicians received and trained medical students from many countries. They also shared instruments and techniques with distant colleagues. In this way laryngology grew as a medical specialism in the second half of the 19th century.

There was also growing interest in diseases of the throat in England at this time. One mid-19th century description captures the British view of quality of observation and interpretation of visualization afforded by the laryngoscope. It comes from George Johnson (1818-1896) Professor of Medicine at Kings College, London in his lectures "The Laryngoscope directions for its use and practical illustrations of its value in the diagnosis and treatment of diseases of the throat and nose" delivered at the Royal College of Physicians. He states: "The larynx being the door-keeper of the lungs, as well as the organ of the voice is largely supplied by nerves and endowed with exquisite sensibility. Its muscular apparatus is therefore readily thrown into a state of spasm, not only by irritation of the larynx itself, but by disturbing

influences transmitted from a distance through the nerves ... before the introduction of the laryngoscope it was often impossible to determine to what extent the symptoms were a result of structural changes within the larynx, and how far they were due to spasm of the laryngeal muscle.”¹⁰

Thus, the development of this instrumentation appears to have driven a great expansion in interest in patients with voice disorder in the late 1850s and 1860s across Europe, as there was now a clinical approach to direct investigation of the throat leading to more precise diagnosis and treatment. As the London clinician Morell Mackenzie said in 1863, “it was not, however till quite recently, when the introduction of the laryngoscope enabled ‘the eye to direct the hand’, that it became possible to apply the galvanic current directly to the nerves and muscles of the larynx”¹². It appears that the development of the laryngoscope directly drove the growth in medical treatment for individuals with voice disorders and a deeper understanding of the function of the larynx in the production of the voice.

Morell Mackenzie’s developing interest in voice disorders

Mackenzie initially trained as a surgeon and was elected a member of the Royal College of Surgeons in 1858. The following year, he went to the Continent to extend his medical training and visited Czermak in Budapest where he learned to use the laryngoscope. This experience had a great impact on him. He returned to London and gained his M.B. in 1861 and his M.D. in 1862 from the University of London. He then took up a post as assistant physician at the London Hospital where he had originally trained¹³. However, his interest in focusing specifically on diseases of the throat, which had been sparked by his time spent with Czermak, motivated him to open his own consulting rooms so that he could concentrate on seeing patients with these problems exclusively. Thus, Mackenzie established the first such institution in London

specifically for this called “The Metropolitan Free Dispensary for Diseases of the Throat and Loss of Voice” at King Street. What Mackenzie had begun single-handedly as a small dispensary soon became the much larger establishment of the Throat Hospital in Golden Square by 1865. In explaining the foundation of this clinic, one motivation Mackenzie pointed to was the development of the laryngoscope which he believed offered new opportunities for diagnosis and treatment.

-----Insert Figure with Mackenzie’s Photo about here-----

From the early 1860s, Mackenzie began to give lectures and publish articles on his chosen specialty, the throat. One of the earliest was “On the application of remedies to the larynx with the aid of the laryngoscope.”¹⁴ He won the Jacksonian Prize essay for the Royal College of Surgeons: “The Pathology and Treatment of Diseases of the Larynx: Diagnostic Indications to include the Appearance as Seen in the Living Person” in 1863. In the same year, he also gave a series of lectures on the “Treatment of Hoarseness” which were published in the *British Medical Journal* and as a book¹⁵. This work primarily dealt with various types of vocal cord paralysis and their treatment with galvanism. This was enlarged and revised in 1868 under the title *Hoarseness, Loss of Voice and Stridulous Breathing*¹⁶. In this book, he recommends the novel terminology which is still in use today: the terms ‘adductor’ to designate the left and right lateral crico-arytenoids and the arytenoideus proprius the central adductor, and ‘abductor’ for the crico-arytenodei postici. There was also a significant addition in the type of voice disorders covered. A new section dealt with “Spasms of the tensors of the vocal chords”. This is where Mackenzie presents his description of what is recognizable as SD.

Mackenzie’s description of SD

Mackenzie begins with a clear definition of the disorder: “spasmodic action of the tensors, causing the vocal cords to be unduly and irregularly stretched, and consequently giving rise to a voice which is feeble, jerky, unsteady, and constantly rising to a high key”¹⁶. As for the etiology, Mackenzie suggests that the cause was not obvious, although several cases were of individuals who had to speak at volume for long periods of time, such as in conversing with deaf relatives.

Mackenzie’s description of the vocal symptoms is so clear, precise, detailed and insightful that it is reproduced here in full:

The sound of the voice is so peculiar in these cases that from it alone they can generally easily be diagnosed. The patient is often able to produce some notes, either in his own natural voice or in a slightly muffled tone; but whilst speaking in this way the current of the voice seems to be partially interrupted, and the sound conveys the idea of an arrested action of the respiratory muscles. In fact, it is very much like the straining and rather suppressed voice of a person engaged in some act requiring the prolonged and steady action of the expiratory muscles (parturition, defecation). After speaking a word or two, or several sentences, in this peculiar tone, the patient may again utter a few words in a comparatively healthy voice, and then may immediately relapse into the diagnostic intonation. No approach to the spasm is perceived as long as the patient whispers, but directly the voice is sounded it becomes apparent. In three cases the spasm was diminished by exertion (such as going upstairs or walking quickly). This appears to me to be due to exhaustion of the expiratory muscles, but perhaps it may be that the quickened circulation caused by the exertion had some beneficial influence on the spasm.¹⁶

While he states that the tense condition of the vocal cords can be easily viewed with the laryngoscope, and points out the differences between this and paralysis of the vocal cords, he suggests that the diagnosis of this disorder can be made simply by listening to the voice. Mackenzie states that “The varying voice of the spasmodic case is so different to the constantly high pitched or totally suppressed voice of the paralytic cases, the laryngoscope is scarcely required for diagnosis”¹⁶. What is even more insightful is his suggestion that “the condition appears to me to be due to some morbid condition of the sympathetic ganglia. The cerebro-spinal system does not appear to be at fault.... it appears evident to me that the affection is not one of simple spasm of the crico-thyroid muscle but one of spasm of ‘the expiratory act’ in which the thoracic and abdominal muscles participate”¹⁶.

With regards to prognosis, Mackenzie is not optimistic and states that it is likely to be both chronic and incurable in most cases. His recommended primary treatment is to insist that on complete vocal rest and only whisper when necessary. External application of belladonna to the throat at night and the inhalation of the fumes of nitrated paper were also suggested, though he admits the action of these applications is not clear: “It is difficult to understand the rationale of the actions of inhalations in this affection, unless the intrinsic muscles are also concerned, but it is possible that where the crico-thyroid is alone morbidly active, the beneficial effect of the inhalation may be due to reflex influence, the primary impression being on the mucous membrane”¹⁶. Eight case reports are included to illustrate this affection which is given the designation Dysphonia from spasm of the tensors of the vocal cords. In the case of a woman patient, Mackenzie significantly stresses that she was not “hysterical”.

While up until this point Mackenzie’s work on vocal disorders in general emphasised to value of the laryngoscope for diagnosis and its use in guiding galvanism as a treatment, it is notable that for this particular voice disorder Mackenzie suggest two things: one that the laryngoscope is unnecessary for diagnosis; and two, that galvanism is not an appropriate

treatment. Mackenzie was typically very energetic in his use of treatments, and his 1868 book is primarily taken over by descriptions of his treatment successes. This is validated in the book review in the *British Medical Journal*¹⁷. However, it is notable that Mackenzie clearly thought that these particular disturbances of vocal cord movement should be treated with rest primarily. This is underscored by his view that the source of the voice disorder is a quite different aspect of the nervous system and that the characterization of the movement was spasmodic rather than spastic. He explicitly states that the difficulty in such patients does not arise from the cortico-spinal pathway, but rather what he refers to as the sympathetic ganglia.

Discussion

The typical citation for the original case of SD is Traube's 1871 patient. This young girl who was suffering from typhoid fever was largely aphonic, producing some falsetto voice. Her voice recovered spontaneously within a matter of days along with other features of her illness. Traube suggested that this "spastic" disorder was a "hysterical" affection. This description does not appear to share many features with what is currently described for SD¹⁸. We argue here that Mackenzie in 1868 offered a description of the clinical entity which includes the primary features of our current formulation of the disorder (See Table). It is notable that although, Türck's student Johann Schnitzler (1835-1893) is credited with coining the term 'Spastic Dysphonia' in 1875, it was Mackenzie who used the denomination of "spasmodic" in his 1868 description of these cases of "dysphonia".

-----Insert Table with comparison about here. -----

With regard to etiology, Traube's single case was suffering from typhus and her voice recovered as her general condition improved. She displayed several other motor signs

involving the tongue and hand. Mackenzie described a number of patients with strained and intermittent voice and interrupted tone, while displaying no other physical symptoms. He observed that the vocal cords appear tense when viewed by the laryngoscope but that diagnosis can be made by the voice quality alone. He makes it clear that this is not a hysterical disorder. Mackenzie recommended vocal rest as a way to treat this chronic problem. This is in contrast to Traube's case which was identified as hysterical and spontaneously recovered. The selection of Traube's historical citation by Luschinger and Arnold, and its propagation throughout the modern literature,³ may be due to the strongly held view by them and subsequent clinicians in the 20th century that the etiology of SD was psychogenic problem and that the appropriate form of treatment was psychiatric.

Mackenzie's view was quite different. In his major textbook, *Manual of Disease of the Throat and Nose* in 1880¹⁹ he discusses "neuroses of motion" which he divided into two natural classes: 1) the loss of power or paralysis; and 2) the "perverted power, or spasm" with two further divisions for spasms of the adductors and spasms of the tensors. His experience of patients with this affection in the 12 years since his original observations in 1868 caused him to remark that that the condition must be rare. Although he perhaps had one of the largest clinics for voice disorders anywhere in the world seeing many tens of thousands of outpatients, he had by then seen only 13 cases with spasm of the tensors of the vocal cords. This is in line with current estimates of prevalence. However, Mackenzie also reports that his were primarily male patients, while today SD is thought to be more than twice as common in women. In his experience, no treatment had afforded a permanent cure, but he believed there was a neuropathology underlying the disorder. Although he had no opportunity to do a post-mortem on a case, he offered the opinion that the pathological changes were too subtle to be detected. Interestingly, although in his comprehensive literature review of others' work

Mackenzie does cite Schnitzler's cases²⁰ as being similar to his own, he does not mention Traube's case.

In one of his much later publications, Mackenzie offers the astute insight that SD shares features with other dystonic movement disorders. While discussing "fatigue of the voice" in his manual on vocal hygiene, he states:

... a still more severe degree of this disorder is what may be called "vocalist's cramp" in which the muscles from over use lose the power of contraction (like an india rubber cord that has been overstretched) or can act only in an irregular and spasmodic way independently of the will or even contrary to it. Analogous conditions are often seen in other parts which have been over taxed, as for instance in the muscles of the hand in persons who write much (scrivener's palsy), or in the muscles of accommodation in the eye in those whose work obliges them to look intently at small objects by artificial light.²¹

It is a puzzle why there has been a general lack of Mackenzie's recognition for his contribution to SD over the decades. In the 1872 revised edition of William Aitken's (1825-1892) bestselling general medical textbook *The Science and Practice of Medicine*, Mackenzie's 1868 book on Hoarseness and his description there of spasms of the vocal cords is reproduced almost verbatim²². This may reflect the high regard for Mackenzie's expertise before his reputation suffered from his involvement in the case of Frederick III²³. It is notable perhaps that a description of SD is included in the second edition of William Gowers' (1845-1915) highly regarded *Manual of Disease of the Nervous System*. However, Mackenzie's work is not mentioned. Gowers describes it as a rare condition analogous to writer's cramp as Mackenzie had previously suggested. However, Gowers classifies it as a functional disorder and only cites Schnitzler's 1875 work and that of several other German authors. This

emphasis on German sources may simply be a reflection the high standing of that country's medical science in this instance²⁴. Nevertheless, acknowledgement of Mackenzie's clinical expertise and scientific acumen cannot be denied. He was likely the most expert clinician on disorders of the larynx in the world at the time. Stevenson enumerates his achievements: the first English laryngologist, founder of the first hospital for diseases of the throat, founder of the *Journal of Laryngology*, and author of the first standard textbook on that specialty²⁵.

We argue that Mackenzie's description of SD in 1868 and further elaborations in 1880 and 1886 contain most of the points which are recognized today in the diagnosis, symptoms, prognosis and treatment. However, there was little recognition of his clinical contribution on SD, perhaps due in part to various negative effects on his reputation in the 1880s. In contrast, his more popular book on vocal hygiene had an incredibly wide circulation which continued for many decades²⁶. We suggest that there is value in revisiting the work of clinicians in the 19th century for insights into current disorders. When writing about SD in our publications we should not continue to erroneously cite Traube's 1871 case of a hysterical girl with typhus as having historical priority. Mackenzie must be credited for the first clear observations on this laryngeal movement disorder.

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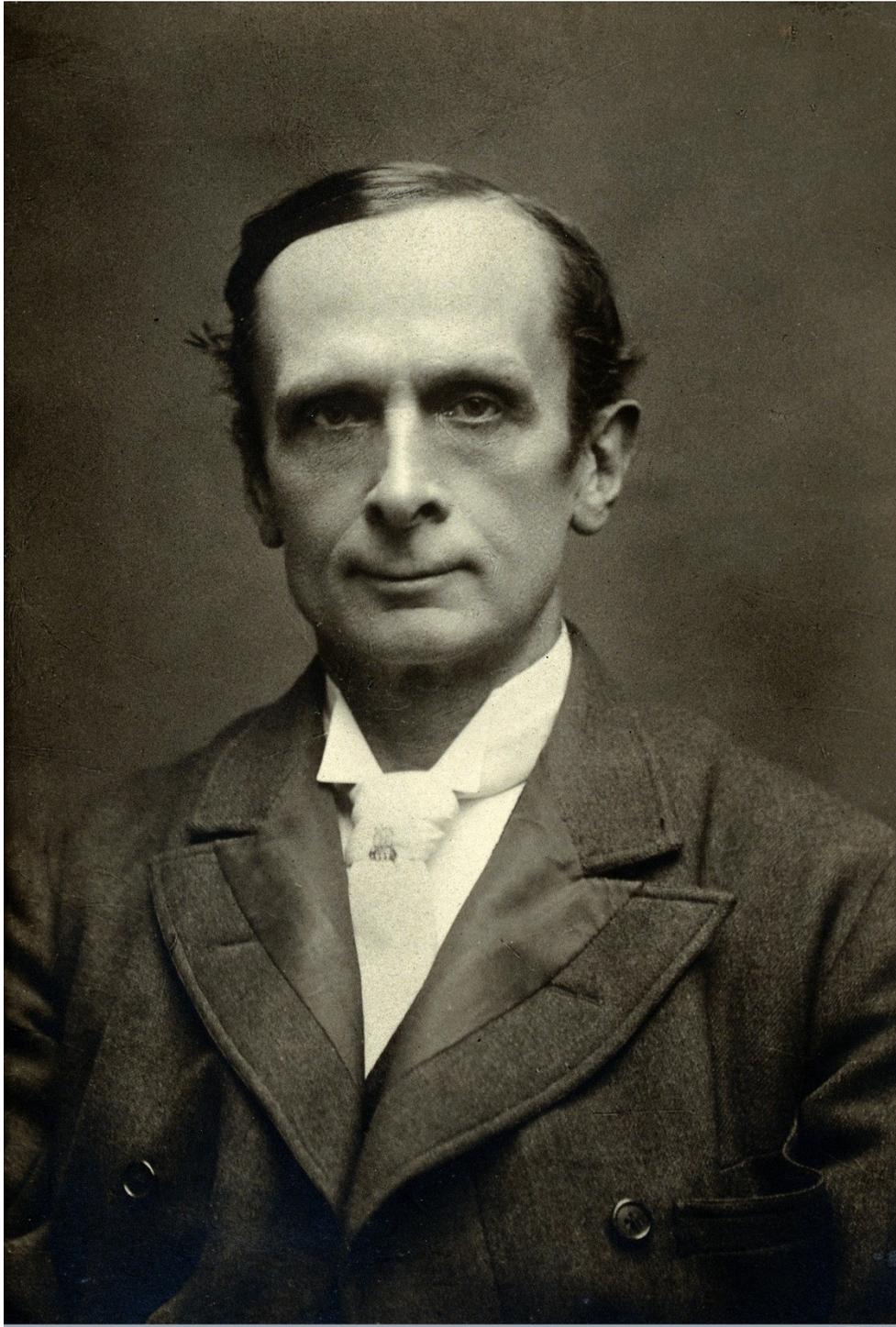


Figure. Sir Morell Mackenzie. Photograph by Elliott & Fry. Credit: Reference No.: ICV No 27226, Iconographic Collection, Wellcome Library, London. Copyrighted work available under Creative Commons Attribution only licence CC BY 4.0 <http://creativecommons.org/licenses/by/4.0/>

Table. Comparison of clinical characteristics of SD from Traube’s original description, Mackenzie’s cases and current understanding.

Clinical Characteristics	Traube One Case, 1864/1871	Mackenzie 13 Cases, 1868 -1880	Current
Age at onset	“Young girl”	30-50 years	30-50 years
Gender	Female	Predominantly Male	Predominantly Female
Incidence	N/A	Rare	Rare
Voice Symptoms	“Aphonic” “with falsetto voice”	“Feeble, jerky, unsteady and constantly rising to a high key”	Strain-strangled voice, phonatory breaks, pitch breaks
Time Course	Recovered within days	Chronic	Chronic
Description of Laryngeal Movement	Spastic	Spasmodic	Spasmodic
Cause	Typhoid Fever	Unknown	May have a genetic component
Antecedent Event	None	Vocal overuse noted in some	A minority of patients identify antecedent throat trauma
Comorbid Symptoms	Protrusion of lips and tongue, Hand tremor	None	Other focal dystonias; Vocal tremor
Pathology	“Hysterical”	“morbid condition of the sympathetic ganglia”	Neurological system control disorder
Prognosis	Acute, Recovered	Chronic, Incurable	Chronic, Incurable
Treatment and Management	Galvanism not suitable	Vocal rest	Intralaryngeal Botulinum toxin A injections