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**Title:** Tracing Spasmodic Dysphonia: the source of Ludwig Traube's priority

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## **Abstract**

Objectives: Since the mid-20<sup>th</sup> century, one citation is given historical priority as the first description of Spasmodic Dysphonia (SD): Ludwig Traube's 1871 case of the "spastic form of nervous hoarseness". Our objective is to understand how this case serves as the foundation of understanding laryngeal movement disorders.

Methods: The original German paper was located and translated. Bibliographical and bibliometric methods are used to determine the citation history of this original source over the past 140 years.

Results: Although secondary citations in contemporary publications typically credit Traube for establishing the clinical entity SD, his case does not conform to currently accepted diagnostic features. Citation patterns indicate the source of Traube's priority is publications by Arnold and Luchsinger, mid-20<sup>th</sup> century ENT clinician, particularly their influential 1965 textbook used to train US and UK clinicians on voice disorders for several generations.

Conclusions: Sometimes secondary citations in medical literature lead to the inadvertent perpetuation of factual misrepresentation. The clinical picture of Traube's original case does not represent what clinicians would recognize as SD today. The rich 19<sup>th</sup> century literature on voice disorders is a valuable resource for present day clinicians.

**Keywords:** Dysphonia, Vocal Cord Dysfunction, Spasmodic Dysphonia, Voice Disorders, Movement Disorders, Bibliometrics

## Introduction

Current research investigating Spasmodic Dysphonia (SD), otherwise referred to as Laryngeal Dystonia, gives priority for the first case description to Ludwig Traube's "Spastische Form der nervösen Heiserkeit" (spastic form of nervous hoarseness).<sup>1</sup> An indication of the widespread acknowledgement of Traube's contribution is evinced by over 70 citations on Google Scholar and 53 on Web of Science for his 1871 publication. The earliest modern publication to use this source is a 1968 paper on "Spastic Dysphonia" by Bicknell and colleagues<sup>2</sup>. The citation of Traube's publication as marking the beginning of SD is perpetuated up to the present day in numerous papers published in 2015<sup>3</sup>.

Although currently, the medical community credits priority to Traube's 1871 German language paper for the description of SD, the source of this citation and the association with a psychogenic etiology can be traced back to a highly influential mid-20<sup>th</sup> century American textbook *Voice, Speech, Language* co-authored by Richard Luchsinger and G.E. Arnold<sup>4</sup>. This is where they state: "The first description of a spastic form of psychogenic hoarseness had been given by Traube in 1871. Four years later, the Viennese laryngologist Schnitzler ... published a detailed account of spastic dysphonia, coining the diagnostic term"<sup>5</sup>. The citation for this point is to a paper by Arnold published in an institutional publication that had a very limited circulation<sup>5</sup>.

We have obtained a copy of Traube's original German publication and have translated it into English. In the course of this investigation, it became apparent that this particular case had been initially published in a weekly medical journal in 1864 by Traube's clinical assistant Dr Hermann Fischer<sup>6</sup>. Fischer's case description was subsequently reprinted in Traube's textbook *Gesammelte Beiträge zur Pathologie und Physiologie* in 1871<sup>7</sup>. In scrutinizing of the

symptoms, diagnosis, and recovery of this patient, it is apparent that several features are at odds with our current clinical picture of SD.

We will argue that the citation practice, which identifies Traube 1871 as the first description of SD, appears to be due to its successful transmission by the German-trained clinicians Luchsinger and Arnold. In order to appreciate the historical propagation of clinical ideas regarding SD, we detail relevant biographical information about the main actors-- Ludwig Traube, G.E. Arnold, and Richard Luchsinger.

This paper explores the tendency for secondary citations in the modern medical literature to lead to misrepresentation and conceptual misunderstandings that become embedded and perpetuated across time. We will consider the descriptions and diagnoses contained in Traube's paper, the representation of it in the mid-20<sup>th</sup> century work by Luchsinger and Arnold, and the subsequent citations of this paper in the growing literature on SD.

### **Traube's 1871 case of 'spastic hoarseness'**

Ludwig (Louis) Traube (1818-1876). Traube was born in 1818 in Ratibor, Oberschlesien, in German-speaking Silesia (Czech Republic). He was trained at the University of Breslau (Wroclaw) and developed his interest in physiology under Jan Evangelista Purkinje (1787-1869) and Johannes Muller (1805-1858)<sup>8</sup>. In 1840, he received his medical doctorate from the University of Berlin for a thesis on pulmonary emphysema. In 1843, Traube returned to Berlin and was appointed "Privatdocent" at the Charité Hospital. One of Traube's close colleagues was Rudolf Virchow (1821-1902), and Traube assisted him in the founding of the *Archiv für*

*pathologische Anatomie und Physiologie und für klinische Medizin* (now known as *Virchow's Archiv*).

Traube gained a growing reputation in Berlin medical circles, and was soon promoted to full Professor. He trained several generations of students, and his clinical teaching skills were widely recognized. For example, the great American clinician William Osler (1849-1919) highly valued his visit to Traube's lab in his postgraduate training tour of Europe<sup>8</sup>. Traube's final three volume work on physiology and clinical medicine<sup>7</sup>, which included the particular case of interest here, was highly esteemed.

Traube's clinical interest in the larynx. Traube's primary clinical interest was actually in pulmonary diseases. Traube is known for coining the term 'metallic sounding wheeze' in asthma and his description of a semilunar space in chest percussion bears the eponym 'Traube's space'. Traube's secondary interest in the larynx was due to its role in various pulmonary diseases (e.g., typhoid fever, diphtheria, tuberculosis, bronchitis, pneumonia, and syphilis) which were common at the time. The objective of his instrumentally based approach was in correlating clinical pulmonary disease with underlying abnormality. To this end, he employed the newly improved laryngoscope to investigate the structure and function of the vocal cords.

In 1864, a brief report from Traube's clinic appeared in the medical press on laryngeal disorders in patients suffering from Typhus. It was authored by Traube's clinical assistant, the Surgeon Dr Hermann Fischer (1830-1919)<sup>6</sup>. Fischer detailed a number of cases and a clinical presentation given by Traube on 24 November (presumably 1863), where he discussed "*nervösen Heiserkeit*" (nervous hoarseness).

Fischer reported that in his clinical demonstration, Traube differentiated between two forms of nervous hoarseness: 'Paralytic' or 'Spastic'. To illustrate the Paralytic form, three cases were described: 1) a patient who had overcome measles in whom the paralysis

disappeared quickly and responded to electrical treatment; 2) a “hysterical” young girl with cramps when swallowing, who had temporary relief from electrical treatment; and 3) a man with an aortic aneurysm and unilateral vocal cord palsy and paralysis of the left inferior laryngeus. For the Spastic form of nervous hoarseness, the observations on one of Traube’s patients is detailed: a “hysterical” young girl. It is this case description for which Traube’s priority for SD is now attributed.

The term ‘hysterical’ must be understood in its historical context. Before the advent of Jean-Martin Charcot’s and subsequently Sigmund Freud’s work on the topic beginning approximately a decade after the period of interest here, the term was used to indicate any medically unexplained symptoms.<sup>9</sup> In the *Oxford English Dictionary* definition of ‘hysterical fever’, it offers a quote from the early 19<sup>th</sup> century: “It [*sc.* mild typhus] has sometimes been denominated the hysterical fever.”<sup>10</sup> Moreover, the term ‘nervous’ also had a meaning that is somewhat different from present usage. In the 19<sup>th</sup> century, the term simply meant relating to the nerves and the nervous system. Nervous diseases were considered to be disorders or dysfunctions arising from the nervous system.

A case of Spastic Nervous Hoarseness. Traube’s patient was described in Fischer’s 1864 report as being very hoarse, and nearly aphonic. She could only intermittently, and with great effort, produce high falsetto voice utterances. Examination with the Laryngoscope showed spasmodic closure of the glottis, whereby the left arytenoid cartilage moved over the right, while the epiglottis was bent steeply to the back. Electrical sporadic current treatment would not be a treatment option in this case. The cause of the nervous hoarseness was attributed to the extreme muscle tension of the vocal cords, accompanied by protrusion of the tongue and lips during speech. There was also hand tremor. All of these symptoms were present from the beginning of the illness. As the patient regained her strength, and her hand tremor and spasms of the lips and tongue reduced, her voice returned over a period of a few days. It was concluded that this

sudden outcome would not have been possible if caused by a structural abnormality. The question was raised whether this type of paralytic hoarseness may be seen in the first week of a typhus infection co-occurring with other symptoms of serious “nervous depression” (i.e., reduced constitutional vitality) as in cases of diphtheria. Fischer’s 1864 report was reprinted verbatim in Traube’s textbook of 1871<sup>1</sup> with the full citation credit given to his junior clinician. It is Fischer’s 1864 case, described as having voice, speech and hand movement difficulties as secondary symptoms of typhus, that has been cited as the first description of SD.

### **Arnold’s and Luchsinger’s role in the attribution of Traube’s priority**

In what appears to be one of the first reference to the case in the modern literature, Arnold identifies Traube’s case as being of “psychogenic” origin and suggests that the earliest historical observations include allusions of the psychiatric concepts of their time<sup>5</sup>. Arnold provides the formulation that SD is the result of “psychic trauma”. This is not surprising as psychoanalytic explanations were commonly offered for many other conditions at this time, such as in Tourette Syndrome<sup>11</sup>. Arnold suggests that because SD represents an advanced psychoneurosis cases should only be treated by a Psychiatrist. Luchsinger and Arnold’s textbook asserts that since SD’s earliest description it has been agreed to have a “psychoneurotic” origin.<sup>4</sup> These statements were perpetuated for many decades. A recent historical review repeats the attribution of a psychosomatic origin for SD to Traube’s case<sup>12</sup>. How this formulation of the disorder may have come about is explored through the contributions of Arnold and Luchsinger.

G.E. Arnold (1914-1989), Godfrey E. Arnold was born in Moravia. He trained under Hermann Gutzmann (1892-1972) as a phoniatician. He was the Director of the Neumann Clinic in Vienna, succeeding Emil Froeschels (1884-1972). He was known for his expertise on traumatic and constitutional disorders of speech and voice. Arnold subsequently emigrated to the USA

after the WWII. He became the clinical director of the National Hospital for Speech Disorders in New York (f. 1916), taking over from the founder Dr. James S. Greene (1880-1950). He went on to establish the division of Otolaryngology at the University of Mississippi at Jackson.

Richard Luchsinger (1900-1993). Richard Luchsinger was born in Switzerland and received medical training in Basel. He was director of the Voice Clinic in Zurich. Luchsinger was one of the founders of the first academic journal on voice, *Folia Phoniatica* (f. 1947), and was the second president of the International Society of Logopedics and Phoniatics (IALP, f. 1924) from 1953-9.

Textbook representations. Luchsinger and Arnold published the *Lehrbuch der Stimm-und Sprachheilkunde* (Textbook of Voice and Speech Habilitation) in 1949<sup>13</sup>. In this German text, Luchsinger authored the section on the Physiology and Pathology of Phonation and Respiration. He discussed spastic dysphonia in a chapter on functional voice disorders and identify Traube as the source of the earliest description. After WWII, one of Arnold's first initiatives at the National Hospital of Speech Disorders was to replace Greene's magazine on stuttering *Talk* with a more scholarly Bulletin entitled *Logos*<sup>14</sup>. In the second volume, Arnold published an article on "Spastic dysphonia: I. Changing interpretations of a persistent affliction."<sup>5</sup> Interestingly, Arnold does not give outright priority to Traube here. He says only that the condition has been known at least since Traube in 1871.

In their co-authored 1965 English textbook<sup>4</sup>, Luchsinger again gave priority to Traube for the earliest description of SD, but the source is now attributed to Arnold's paper rather than their original German text<sup>5</sup>. Interestingly, Arnold's 1959 paper is the source of over 70 citations in the SD literature according to Google Scholar, even though it was published in an institutional bulletin with a small circulation and it is now relatively difficult to find a copy.

For example, it is referenced as the source of Traube's priority in a widely cited current textbook on voice disorders<sup>15</sup>, and is also cited in recent research papers on SD<sup>16</sup>.

There appears to have been a hiatus in research on SD in the first half of the 20<sup>th</sup> century. According to one of the early modern papers on SD from the 1960s<sup>2</sup>, spastic dysphonia was found to be rarely mentioned in neurological texts and journals, although known by laryngologists for nearly a hundred years. After this comment, they provide Traube 1871 as the citation. The authors go on to quote Arnold's point that the history of the term 'dysphonia spastica' is a typical example of the repeated re-discovery of a previous well-known condition and the invention of a new name to replace an old one, although the conditions are indistinguishable. We suggest that the case from Traube's clinic does include features that distinguish it from present day descriptions of the condition. This is not merely an instance of changes in nomenclature, nor can it be attributed to improved understanding brought on by imaging or innovations in genetics. Rather, it is a reflection of the different cultural and clinical context in which these investigations were being pursued.

## **Discussion**

Although there has been intensive work on the nosological refinement of SD from the second half of the 20<sup>th</sup> century to those used today, the citation of Traube 1871 continues to be given to mark the beginning of research into laryngeal movement abnormalities. The original case was of a spastic form of nervous hoarseness in a young woman whose voice disorder was the result of typhus that recovered in a matter of days published by Fischer in 1864. Our investigation has traced the source of the current citation practice that gives this case priority for spasmodic dysphonia to the influential contributions of Arnold and Luchsinger in the mid-20<sup>th</sup> century. Moreover, no other citation is offered from the historical literature before 1871. There was in

fact a great deal of activity in the investigation of laryngeal disturbances, once the laryngoscope came into widespread clinical use in the 1860s throughout Europe.

The British ENT clinician Felix Semon (1849-1921) wrote an extensive literature review of the investigation of laryngeal paralysis in 1892. Semon documents the large number of articles that had already appeared on this subject before 1871. He stated, "During the years 1863 to 1876, a large series of valuable communications were published, which increased our knowledge concerning the individual forms of laryngeal paralysis." Semon continues commenting "...without however, formulating any general principles concerning the laws governing the production of these different forms."<sup>17</sup> Interestingly, it should be noted that in this review Semon also gave priority to Traube for his description of laryngeal paralysis: "The distinction of having diagnosed the first laryngeal paralysis by means of the laryngoscope belongs, so far as I know, to my illustrious teacher, L. Traube."<sup>17</sup> In Semon's conclusion to his review of laryngeal paralysis he states,

"I conclude with expressing the wish that in the further development of the subject we may be spared any further attempt at founding a special laryngeal pathology; and that it may be always remembered that the true scientific course is to refrain from premature explanations of observed facts. The true causation must eventually be reached by quietly and patiently following up and carefully comparing observations. He who is led away by mere ingenuous views, like the man who follows a Will-o'-the-Wisp, loses time and often lands in a morass."<sup>17</sup>

Semon seems to offer sound advice, which could fruitfully be applied to the characterization of spasmodic afflictions as well.

## **Conclusion**

If, in the 20<sup>th</sup> and 21<sup>st</sup> century, subsequent researchers had carefully read the actual case rather than the assertion by Luchsinger and Arnold, it would have been readily apparent that what was described was not that of SD, and thus the years of misattributing a psychosomatic origin to this disorder may have been avoided. A number of points have been determined by the translation and investigation of the original publication. Two minor bibliographic facts of record need to be acknowledged. Firstly, the case was published in 1864, not 1871. Secondly, the author citation should be credited to Fischer not Traube, although the case did come from his clinic. More significantly the clinical and diagnostic facts about the nature of this case are also quite different from what secondary sources suggest. Firstly, the description of the vocal cords and the voice disorder indicate the patient was suffering a temporary aphonia due to sustained muscle tension. Secondly, there was no suggestion that the etiology was psychogenic, but as a consequence of a severe generalized illness. This young girl suffered from typhoid fever, and briefly exhibited a speech production difficulty which resolved as her general health improved.

Semon's extensive review at the end of 19<sup>th</sup> century on laryngeal paralysis cited clinical and experimental research from sources in multiple languages and countries. The evidence presented here shows a contrasting picture of research scholarship at the beginning of the 21<sup>st</sup> century regarding SD. From our investigations of citation patterns in this literature, one German paper reprinted six years after its initial appearance that describes a patient whose clinical characteristics do not match current descriptions of SD, is typically the only citation offered for historical work on this disorder. It appears to be not only a misattribution of priority but also to indicate a seriously limited awareness of the rich store of 19<sup>th</sup> century clinical research.

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