Complex regional pain syndrome related movement disorders: studies on pathophysiology and therapy.

Munts, A.G.

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How psychogenic is dystonia? Views from past to present

Alexander G. Munts, MD,¹,² and Peter J. Koehler, MD, PhD, FAAN³

¹Department of Neurology, Kennemer Gasthuis, Haarlem, The Netherlands
²Department of Neurology, Leiden University Medical Centre, Leiden, The Netherlands
³Department of Neurology, Atrium Medical Centre, Heerlen, The Netherlands

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Abstract
In the last few centuries there has been a constant sway between organic and psychogenic explanations for dystonia. In the current study we investigate this history, assuming the perspective of a spectrum from organic to psychogenic, between which ideas were moving. We have focussed on (i) primary generalised dystonia; (ii) cervical dystonia; (iii) writer’s cramp; and (iv) fixed dystonia related to complex regional pain syndrome. We have studied medical texts published since the 19th century and their references. Jean-Martin Charcot advocated the concept of hysteria: disorders in which, besides predisposition, environmental factors were involved in its pathogenesis. Sigmund Freud introduced psychoanalysis as an explanatory therapy for psychic disorders. Previous theories, together with the lack of an organic substrate for dystonia, made a strong case for psychogenic explanations. Consequently, many dystonia patients were told that they suffered from psychological conflicts and were treated for them. However, after the description of new hereditary cases in the 1950s, the limited efficacy of psychotherapy in torsion dystonia, the effects of surgical treatments and the lesion studies in the 1960s, more physicians became convinced of the organic nature. The culminating point was the discovery of the DYT1 gene in 1997. In the meantime, experts had already convinced the neurological community that cervical dystonia and writer’s cramp were focal dystonias, i.e. minor forms of generalised dystonia, and therefore organic disorders. In contrast, the pathophysiology of fixed dystonia related to complex regional pain syndrome remained controversial. Knowledge of this history, which played on the border between neurology and psychiatry, is instructive and reflects the difficulty in discriminating between them. Today, new insights from functional imaging and neurophysiological studies again challenge the interpretation of these disorders, while the border between psychogenic and organic has become more blurred. Abnormalities of sensorimotor integration and cortical excitability that are currently supposed to be the underlying cause of dystonia bring us back to Sherringtonian physiology. We suggest that this may lead to a common explanation of the four afflictions of which we have traced the history.
Introduction

For many years, physicians have observed and discussed the remarkable signs of what we nowadays call dystonia. The introduction of the term dystonia as an abnormality of tone with coexistent hypo- and hypertonia goes back to 1911 when the well-known Berlin neurologist Hermann Oppenheim (1858-1919) introduced *dystonia musculorum deformans*, which was later renamed early-onset generalised torsion dystonia. In 1967, Wolfgang Zeman (1921-2001) and Paul Dyken reported the presence of milder forms of dystonia in dystonia musculorum deformans families, including cases of isolated writer's cramp. In 1976, David Marsden (1938-1998) proposed the term focal dystonia for blepharospasm, oromandibular dystonia, dystonic writer's cramp, and torticollis, as well as for axial dystonias, arguing that these were closely related to generalised dystonia. Up to the present, this view has not changed. Over the years, however, there has been a discussion on whether the aetiology of dystonia is either organic or psychogenic. In this paper we study the evolution of ideas with respect to dystonia, in particular whether or not it was considered an organic or psychogenic affliction. We will put the historical evolution against the background of present-day knowledge resulting from functional imaging and neurophysiological studies, and of the blurred border between organic and psychogenic.

Methods

We started our search on dystonia history using two standard books on the history of medicine. Furthermore, we used the PubMed database by entering the term 'dystonia' with limitation to 'history of medicine'. In addition, we used medical and neurological textbooks from the 19th and 20th century written in English, French, German, or Dutch. In the tables of contents and subject indexes we searched for dystonia, spasm(s), spasmodic contortion or contraction, torticollis, wryneck, (writer's) cramp, scrivener's palsy, occupational neurosis (English); dystonie, torticolis (mental), spasme clonique (du sterno-mastoïdien), spasme fonctionel (du sterno-mastoïdien), crampe fonctionnelle, crampe des écrivains (French); Dystonie, Torticolis, Schreib(ek)rampf, Funktionskrämpfe, Beschäftigungsneurose (German); dystonie, torticolis, (ver)krampf(ing), and schrijverskramp (Dutch). In addition, we searched for relevant literature in the reference lists of consulted books and papers. As many nineteenth- and early twentieth-century primary textbooks refer to the work of Duchenne and Bell, we chose to discuss their descriptions...
in more detail. When dealing with the question whether a particular author considered a disorder psychogenic or organic, we assumed a spectrum from organic to psychogenic between which ideas of the individual authors could be placed, as far as could be derived from the text.

**Definition of dystonia**
The word *dystonia* was introduced in 1911. Later its meaning was changed several times. For example, Derek Denny-Brown (1901-1981) considered dystonia a disorder with a fixed posture or oscillation between two or more fixed postures (Denny-Brown, 1965; Denny-Brown, 1966). The modern definition is "a syndrome of sustained muscle contractions, frequently causing twisting and repetitive movements, or abnormal postures". In this article, we use the latter definition.

**Definitions of neurosis and hysteria**
In the late 18th and early 19th century, neurosis was defined as the category of clinically well-characterised nervous diseases without known pathological substrates. Throughout the 19th century, this category became smaller when neuropathological substrates of several of these diseases were established. Hysteria was a subcategory within the neuroses, in which neurological signs were similar to those in patients who suffered from nervous diseases with known anatomic lesions, although somewhat different and usually more extensive. In the late 19th century, Charcot assumed that hysteria arose from a lesion of an undetermined structural or functional nature and he expected that the pathological basis would be found in due course. The neurological defect was believed to result from a combination of hereditary predisposition and an environmental, provocative factor, which usually was a physical or emotional shock. Therefore, throughout history the term psychogenic cannot always be considered equal to non-organic, in particular in the pre-Freudian period. After this period non-organic mostly did mean psychogenic. The meanings of neurosis and hysteria changed and finally the terms were used solely in descriptions of psychiatric diseases. At present, the terms are used less often, and no longer listed in the Diagnostic and Statistical Manual of Mental Disorders. In this article, where needed, we clarify the context of these words.
Results

Primary generalised dystonia

In 1871 William Hammond (1828-1900) reported on three patients "in which the most characteristic symptoms are an inability to retain the fingers and toes in any position in which they may be placed, and their continual motion". He mentioned the acquired disorder 'athetosis' and hypothesised on a striatal lesion. Hammond's patients may not be considered dystonic patients – although today most authors consider athetosis part of the dystonia spectrum but it is important to mention Hammond's coining of the term athetosis. In 1897, the Spanish physician Lluis Barraquer I Roviralta (1855-1928) reported another patient with athetosis, which later was considered the first description of generalised torsion dystonia. In 1908, the German Marcus Walter Schwalbe (1883-1927) described hysterical symptoms in the siblings Fanny, Heimann, and Wulf Levin, suffering from tonic cramps, which is now recognised as early-onset generalised torsion dystonia (Figure 2.1).

Among the most important hysterical characteristics there was the presence of pressure points (called 'hysterogenic zones' in Charcot's work), i.e. body areas in which cramps may be provoked by pressure. Familial involvement was another feature. In 1911 Oppenheim launched the term dystonia musculorum deformans for the same disorder. He reported on four patients, who were Jewish children. Illustrative is the description of a 14-year old girl with a 'dromedary gait' "indem der sättelformige ausgebuchtete Rücken in eine fast horizontale Lage kommt, und zwar fällt die Rumpfbeugung zusammen mit dem Aufsetzen des linken Beins, während der Rumpf sich hebt beim Schwingen des linken Beins" [because the saddle-shaped back acquires an almost horizontal position, in which the left leg posture phase is accompanied by trunk flexion, and the swing phase by trunk elevation]. He was convinced that it was an organic disease without concomitant hysteria.

Subsequently, dystonia musculorum deformans became a collective term for a variety of neurologic disorders. There was a continuing discussion on the characteristics of the disorder, and a pathological substrate was still unknown. For these reasons, the concept of dystonia as a disease was demolished during the tenth Réunion Neurologique Internationale Annuelle in Paris (1929). Subsequently, the Danish physician Auguste Wimmer (1872-1937) concluded that dystonia was no more than a syndrome. In the meantime, a psychogenic explanation had emerged for various nervous disorders without anatomic lesions. One of the founders of psychogenesis was Sigmund Freud (1856-1939). From 1888 to 1910 he described several patients who suffered from hysteria and in whom
symptoms were related to conflicts and psychological defence.\textsuperscript{31} The effectiveness of psychological intervention supported this new and revolutionary theory. Exploring and resetting the unconscious mind, by means of 'psychoanalysis', became a successful therapy in many cases of hysteria. Since then, many patients with generalised dystonia underwent this or other forms of psychotherapy.

In 1944, Ernst Herz (1900-1965) published three frequently cited articles on his studies of dystonia cases. He considered dystonia a "clinical entity" with "characteristic irregular, involuntary motor phenomena", "a peculiar distribution of 'excess of motion' and 'excess of tension'", and "without recognizable etiologic factors at onset".\textsuperscript{32,34} In 1959 the hereditary nature of dystonia musculorum deformans was demonstrated\textsuperscript{35} and ten years later a report was published on the limited efficacy of psychotherapy in 44 patients with torsion dystonia.\textsuperscript{36} In the same year, Irving Cooper (1922-1985) reported on a 77% success rate after unilateral or bilateral surgery of the thalamus or globus pallidus in 144 dystonia musculorum deformans patients.\textsuperscript{37} In the 1960s Denny-Brown reported his landmark studies on dystonia. He caused selective lesions in monkey brains which led to uncontrollable abnormal postures and movements resembling dystonia. It was remarkable

\textbf{Figure 2.1.} Developments on generalised dystonia in the 20\textsuperscript{th} century. See the text for references.
Not for the first time but more convincing than ever before.
to observe that damage in different anatomical structures might have the same consequences. Denny-Brown assumed that dystonia resulted from an imbalance of reflex responses in the central nervous system. \(^{38-40}\) In 1975, an International Symposium on Dystonia was organised. In the preface of the conference book, Eldridge and Fahn wrote:

> In the past, many victims of dystonia and their families have been caused anguish and hardship over and above that caused by the disease itself owing to the frequent misdiagnosis of the symptoms as manifestations of a psychiatric ailment. We hope that the present volume will facilitate accurate diagnosis, assist practicing physicians in treating their dystonic patients, encourage them to report their observations and results, and stimulate clinical and basic research workers in efforts to elucidate the causes and eventual treatment of dystonia and related disorders. \(^{41}\)

At this symposium, Marsden emphasised the existence of sporadic torsion dystonia. \(^{42}\) Fahn and Eldridge stated that psychologically based dystonia was a rare or non-existent condition. \(^{43}\) However, three years after the symposium (1978), the "first case of psychogenic dystonia" was reported \(^{44}\) and in 1983, at the 35\(^{th}\) annual meeting of the American Academy of Neurology, another five followed. \(^{45}\) The first patient was a 15-year-old girl who had simulated her dystonic symptoms and signs. She was admitted after a failed suicide attempt and told that she had faked her symptoms: "she discarded her leg brace, and the sustained contractions in her leg and arm immediately improved". The histories of the other five patients were not included in the publication.

In 1984, an ad hoc committee, consisting of members of the Scientific Advisory Board of the Dystonia Medical Research Foundation, re-defined dystonia as "a syndrome of sustained muscle contractions, frequently causing twisting and repetitive movements, or abnormal postures". \(^{17}\) Four years later a classification for psychogenic dystonia followed (Table 2.1). \(^{46}\) The first locus (9q32-34 region) for idiopathic dystonia (DYT1) was found in 1989 \(^{47}\) and ten years later the same group identified the gene, describing a unique 3-base pair deletion in the coding region, which was responsible for almost all their cases with early-onset, but for only a few with late-onset idiopathic torsion dystonia. \(^{48}\)
Table 2.1. Definitions on the degree of certainty of the diagnosis of a psychogenic dystonia.\textsuperscript{46}

| Documented | Persistent relief by psychotherapy, by the clinician utilizing psychological suggestion including physiotherapy, or by administration of placebos (again with suggestion being a part of this approach), or the patient must be witnessed as being free of symptoms when left alone supposedly unobserved |
| Clinically established | The dystonia is inconsistent over time or is incongruent with classical dystonia, plus at least one of the following features: |
| | - other neurologic signs are present that are definitely psychogenic, e.g. false weakness, false sensory findings, and self-inflicted injuries |
| | - multiple somatizations are present |
| | - an obvious psychiatric disturbance is present |
| Probable | |
| | - The dystonia is inconsistent over time or is incongruent with classical dystonia, but there are no other features, or |
| | - The dystonia is consistent and congruent with organic dystonia, however at least one of the following features is present: |
| | - other neurologic signs are present that are definitely psychogenic, e.g. false weakness, false sensory findings, and self-inflicted injuries |
| | - multiple somatizations are present |
| Possible | The dystonia is consistent and congruent with organic dystonia, however, an obvious emotional disturbance is present |

**Cervical dystonia (Table 2.2)**

One of the earliest descriptions of cervical dystonia was given by the Swiss physician Felix Platerus, also known as Plater (1536-1614).\textsuperscript{49,50} He described a case of 'spasmi species, in qua caput in sinistrum latus torquebatur' [a kind of spasm in which the head was turned to the left side]. The Dutch Nicolaas Tulp, or Tulpius (1593-1674), well-known from the famous Rembrandt painting *The Anatomy Lesson* of 1632, described dissection of the sternocleidomastoid muscle as a therapy for what he called 'obstipi capitis' [crooked head] in his *Observationes medicae*.\textsuperscript{51,52} However, this patient had had torticollis from childhood and the origin was probably mechanical.
Table 2.2 (continued on next pages). Historical descriptions on cervical dystonia

<table>
<thead>
<tr>
<th>Year</th>
<th>Author</th>
<th>City</th>
<th>Terminology</th>
<th>O/P</th>
<th>Comments</th>
</tr>
</thead>
<tbody>
<tr>
<td>1614</td>
<td>Plater</td>
<td>Basel (CH)</td>
<td>spasmi species, in qua caput sinistrum latus torquebatur</td>
<td>O</td>
<td>case report; explained as a disorder of the muscle and surrounding tissue</td>
</tr>
<tr>
<td>1641</td>
<td>Tulp</td>
<td>Amsterdam (NL)</td>
<td>obstipi capitis</td>
<td>O</td>
<td>probably mechanic origin; dissected the involved muscle</td>
</tr>
<tr>
<td>1765</td>
<td>Lorry</td>
<td>Paris (FR)</td>
<td>colli singularem omninō distortionem</td>
<td>P</td>
<td>case report in a monography on melancholia; explained as due to boredom and</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>therefore aversion to life</td>
</tr>
<tr>
<td>1768</td>
<td>Boissier</td>
<td>Montpellier (FR)</td>
<td>obstipitas spasmodica</td>
<td>O</td>
<td>classified as partial tonic spasms, together with strabismus, tics,</td>
</tr>
<tr>
<td></td>
<td>de Sauvages</td>
<td></td>
<td></td>
<td></td>
<td>contractures, ankylosis, cramps and priapism</td>
</tr>
<tr>
<td>1822</td>
<td>Dupuytren</td>
<td>Paris (FR)</td>
<td>torticolis, caput obstipum</td>
<td>O</td>
<td>divided the sternocleidomastoid muscle</td>
</tr>
<tr>
<td>1825</td>
<td>Middlesex Hospital</td>
<td>London (UK)</td>
<td>spasmodic affection of the muscles of the neck</td>
<td>U</td>
<td>case report</td>
</tr>
<tr>
<td>1825</td>
<td>Gilby</td>
<td>Bristol (UK)</td>
<td>contraction of the muscles of the neck</td>
<td>O?</td>
<td>efficaciously used electricity in the corresponding contralateral muscles</td>
</tr>
<tr>
<td>1838</td>
<td>Stromeyer</td>
<td>Hannover (DE)</td>
<td>Krampf des Kopfnickers</td>
<td>O</td>
<td>dissected the involved muscle(s)</td>
</tr>
<tr>
<td>1844</td>
<td>Bell</td>
<td>Edinburgh (UK)</td>
<td>spasmodic contortion of the head and neck</td>
<td>O</td>
<td>suspected a diseased nerve</td>
</tr>
<tr>
<td>1846</td>
<td>Romberg</td>
<td>Berlin (DE)</td>
<td>Halsmuskelkrampf</td>
<td>U</td>
<td>in most cases unknown cause, sometimes due to physical strain; described</td>
</tr>
</tbody>
</table>

Historical study of dystonia | 27
<table>
<thead>
<tr>
<th>Year</th>
<th>Author</th>
<th>Location</th>
<th>Description</th>
<th>Notes</th>
</tr>
</thead>
<tbody>
<tr>
<td>1861</td>
<td>Duchenne</td>
<td>Paris (FR)</td>
<td>spasme du sternomastoidien</td>
<td>O due to intense emotions may be cured by continuous stretch to the antagonists by means of an apparatus; no success with electricity</td>
</tr>
<tr>
<td>1867</td>
<td>Middlesex Hospital</td>
<td>London (UK)</td>
<td>spasmodic contraction of cervical muscles</td>
<td>O? case report; spinal accessory nerve was dissected, although without efficacy</td>
</tr>
<tr>
<td>1872</td>
<td>Jaccoud</td>
<td>Paris (FR)</td>
<td>hyperkinésie de l'accessoire de Willis</td>
<td>O clonic form: rare, unknown cause; tonic form: either congenital, vertebral disorder or due to pressure on sensible nerve (reflex cramp)</td>
</tr>
<tr>
<td>1873</td>
<td>Charing Cross Hospital</td>
<td>London (UK)</td>
<td>clonic torticollis</td>
<td>O case report; improvement by electricity together with rhythmical exercise</td>
</tr>
<tr>
<td>1888</td>
<td>Charcot</td>
<td>Paris (FR)</td>
<td>spasme clonique du sternomastoidien et du trapèze</td>
<td>O = P case report; improvement with electricity to the atrophied contralateral muscle</td>
</tr>
<tr>
<td>1888</td>
<td>Gowers</td>
<td>London (UK)</td>
<td>spasmodic wry-neck</td>
<td>O + P 'not-organic' variant (= hysterical = partly moral, partly physical) tends to spread from the neck to the trunk</td>
</tr>
<tr>
<td>1889</td>
<td>Freud</td>
<td>Vienna (AT)</td>
<td>Genickkrämpfe</td>
<td>O case report (Frau Emmy v. N...); hysteria patient who underwent hypnosis; Genickkrämpfe were not considered hysterical</td>
</tr>
<tr>
<td>1890</td>
<td>Keen</td>
<td>Philadelphia (US)</td>
<td>spasmodic wry neck</td>
<td>U divided the dorsal rami of the C1-C3 spinal nerves in a patient in whom...</td>
</tr>
<tr>
<td>Year</td>
<td>Author</td>
<td>City/Country</td>
<td>Diagnosis</td>
<td>Nature</td>
</tr>
<tr>
<td>------</td>
<td>--------</td>
<td>--------------</td>
<td>-----------</td>
<td>--------</td>
</tr>
</tbody>
</table>
| 1893 | Brissaud | Paris (FR) | torticollis mental | P | spinal accessory nerve division was unsuccessful; some improvement 
|       |         |              |           |       | believed that torticollis was a tic (P), not a spasm (O); the touch 
|       |         |              |           |       | that was able to correct proved the psychic nature 
|       |         |              |           |       | case report; cured with suggestion 
|       |         |              |           |       | during hypnosis 
| 1894 | Voisin | Paris (FR) | torticollis intermittent | P | 
| 1894 | Oppenheim | Berlin (DE) | Krämpfe im Bereich der Halsmuskeln | O = P | 
| 1896 | de Quervain | La-Chaux-de-Fonds (CH) | torticollis spasmodique | O? | 
| 1900 | Babinski | Paris (FR) | torticollis spasmodique torticollis mental | O | 
| 1902 | Meige & Feindel | Paris (FR) | torticollis mental | P | the 'geste antagoniste efficace' is characteristic; careful 
|       |         |              |           |       | and prolonged observation is needed to distinguish it from 
|       |         |              |           |       | 'torticollis-spasme' (O) 
|       |         |              |           |       | geste antagoniste was named "Brissauds Handgriff" 
| 1905 | Kollarits | Budapest (HU) | torticollis mentalis | P | 
| 1907 | Curschmann | Mainz (DE) | spasmodischen torticollis | O | vestibular disorder; quinine was efficacious 
| 1914 | Mohr | Koblenz (DE) | Torticollis mental | O < P | often in neuropathic patients; organic 
|       |         |              |           |       | causes must be excluded (ocular, auricular; cervical 
<p>|       |         |              |           |       | spine, or brain |</p>
<table>
<thead>
<tr>
<th>Year</th>
<th>Author(s)</th>
<th>Institution(s)</th>
<th>Diagnosis</th>
<th>Approach</th>
</tr>
</thead>
<tbody>
<tr>
<td>1914</td>
<td>Clark</td>
<td>New York Neurological Society (US)</td>
<td>mental torticollis</td>
<td>P</td>
</tr>
<tr>
<td></td>
<td>Wartenberg</td>
<td>Freiburg im Breisgau (DE)</td>
<td>Torticollis</td>
<td>O &gt; P</td>
</tr>
<tr>
<td>1923</td>
<td>Cushing</td>
<td>Boston (US)</td>
<td>spasmodic torticollis</td>
<td>U</td>
</tr>
<tr>
<td>1935</td>
<td>Yaskin</td>
<td>Pennsylvania (US)</td>
<td>spasmodic torticollis</td>
<td>O &lt; P</td>
</tr>
<tr>
<td></td>
<td>Critchley</td>
<td>London (UK)</td>
<td>spasmodic torticollis</td>
<td>O or P</td>
</tr>
<tr>
<td>1940</td>
<td>Kinnier Wilson</td>
<td>London (UK)</td>
<td>torticollis</td>
<td>O or P</td>
</tr>
<tr>
<td>1943</td>
<td>Patterson and Little</td>
<td>Ann Arbor (US)</td>
<td>spasmodic torticollis</td>
<td>O &gt;&gt; P</td>
</tr>
<tr>
<td>1945</td>
<td>Paterson</td>
<td>Edinburgh (UK)</td>
<td>spasmodic torticollis</td>
<td>O or P</td>
</tr>
<tr>
<td>1949</td>
<td>Herz and Glaser</td>
<td>New York (US)</td>
<td>spasmodic torticollis</td>
<td>O</td>
</tr>
</tbody>
</table>

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<table>
<thead>
<tr>
<th>Year</th>
<th>Author(s)</th>
<th>Location</th>
<th>Type of Dystonia</th>
<th>Treatment/Findings</th>
</tr>
</thead>
<tbody>
<tr>
<td>1965</td>
<td>Denny-Brown</td>
<td>Boston (US)</td>
<td>torticollis</td>
<td>influenced by psychogenic factors performed experiments in monkeys; described that torticollis arises from damage to the pretectal region, and is due to distortion of optokinetic reflexes.</td>
</tr>
<tr>
<td>1967</td>
<td>Brierley</td>
<td>Newcastle-upon-Tyne (UK)</td>
<td>spasmodic torticollis</td>
<td>performed experiments in monkeys; described that torticollis arises from damage to the pretectal region, and is due to distortion of optokinetic reflexes.</td>
</tr>
<tr>
<td>1971</td>
<td>Mitscherlich</td>
<td>Düsseldorf (DE)</td>
<td>spasmodic torticollis</td>
<td>efficaciously used behaviour therapy with conditioning through electric shocks.</td>
</tr>
<tr>
<td>1974</td>
<td>Brudny et al</td>
<td>New York (US)</td>
<td>torticollis</td>
<td>P or P = specifically studied the efficacy of psychoanalytical treatments in 60 patients; &gt;5,000 hours; severe ego-regression in all cases.</td>
</tr>
<tr>
<td>1976</td>
<td>Marsden</td>
<td>London (UK)</td>
<td>torticollis</td>
<td>O = organic; P = psychogenic; O &gt; P = majority of cases is organic; O or P = cause is organic in some cases and psychogenic in other cases; O + P = cause is a combination of organic and psychogenic factors in every; U = unknown cause.</td>
</tr>
<tr>
<td>1985</td>
<td>Tsui et al</td>
<td>Vancouver (CA)</td>
<td>spasmodic torticollis</td>
<td>O = organic; P = psychogenic; O &gt; P = majority of cases is organic; O or P = cause is organic in some cases and psychogenic in other cases; O + P = cause is a combination of organic and psychogenic factors in every; U = unknown cause.</td>
</tr>
<tr>
<td>1987</td>
<td>Rentrop and Straschill</td>
<td>Berlin (DE)</td>
<td>spasmodic torticollis</td>
<td>O + P = stated that in some cases psychotherapy is indicated.</td>
</tr>
</tbody>
</table>

O = organic; P = psychogenic; O > P = majority of cases is organic; O or P = cause is organic in some cases and psychogenic in other cases; O + P = cause is a combination of organic and psychogenic factors in every; U = unknown cause.

The well-known Scottish surgeon-anatomist Charles Bell (1774-1842) stated that the origin of 'spasmodic contortion of the head and neck' was nerve rather than muscle dysfunction. One of his patients, Mary Preston, developed the disease following a hard and protracted labour. In Bell's view, a disorder of the accessory nerve but not other...
nerves, due to strain, might lead to unbalanced muscle drive and thus to the disease. Interestingly, the same case was also reported elsewhere,\textsuperscript{57} but according to these authors, the disease was not limited to the distribution of the accessory nerve. Their disagreement with Bell was underlined by their commentary "We have frequently had occasion to notice the very ingenious manner in which Mr. Bell perverts facts, in order to meet his own particular views of a case". The French physician Guillaume-Benjamin Duchenne (1806-1875), who applied electricity for a variety of disorders in Paris hospitals, reported that 'spasme fonctionnel du sterno-mastoidien' is quite resistant to treatment.\textsuperscript{62} Instead, he advised therapy by applying continuous stretch of the antagonists with the use of an instrument.

During one of his well-known Tuesday Lessons, on June 26, 1888, Charcot presented a 63-year old man with 'spasme clonique du sterno-mastoïdien et du trapèze' [clonic spasm of the sternocleidomastoid and the trapezius muscles], which had been present for eight months.\textsuperscript{66} The disorder started after the patient, who was a stockbroker, had lost all his money. On July 10, 1888, he was presented again after being successfully treated with electricity. Five years later, one of Charcot's former students, Edouard Brissaud (1852-1909), introduced the term 'torticollis mental'.\textsuperscript{70} In his view, the condition was psychogenic, which was evident from the fact that the patient was able to correct the powerful muscle activity by simply touching the head, later named the 'geste antagoniste efficace'.\textsuperscript{74} Emphasizing the psychogenic nature again, the Hungarian Jenö Kollarits (1870-1940) reported on six 'torticollis hystericus' cases in 1905. Therapeutic dissection of the involved muscles or nerves, as performed by the Swiss surgeon Fritz de Quervain (1868-1940),\textsuperscript{72} was considered malpractice according to Kollarits, who, instead, stated that therapy should be based on suggestion.

In this period, there was much discussion on torticollis in the scientific community. At the New York Neurological Society (1914), Pierce Clark (1870-1933) presented an adult man with 'mental torticollis' which, he said, was the consequence of pleasurable stroking movements by his mother, before the age of six.\textsuperscript{78} In reaction, Bernard Sachs (1858-1944) said, "if this indicated the future trend for our present-day neurology, then the less we hear of it, the better". The debate went on for several decades. In 1935 Joseph Yaskin (1891-1955) wrote that before surgery, every case of 'spasmodic torticollis' should receive a trial of psychotherapy.\textsuperscript{81} In 1943 Patterson and Little reported on 103 cases with spasmodic torticollis,\textsuperscript{83} stating that the aetiology was usually organic and that surgery, intradural rhizotomy in particular, was very satisfactory. However, in 1945 the Scottish
physician Paterson presented 21 cases, concluding that psychotherapy was the treatment of choice unless gross signs of neurological disease were present.84 At the 1975 International Symposium on Dystonia (vide supra), Marsden presented arguments that spasmodic torticollis, as well as blepharospasm, oromandibular dystonia and dystonic writer's cramp (vide infra), were focal dystonias with an organic aetiology. He summarised the reasons why they had been regarded psychogenic (Table 2.3),3 and subsequently explained his ideas about functional abnormalities in the extrapyramidal motor system.3 Obvious arguments were their occurrence in early-onset generalised torsion dystonia and the similarities with late-onset generalised torsion dystonia: both focal dystonia and late-onset generalised torsion dystonia had a comparable age of onset and were usually neither progressive nor hereditary. A new name was introduced in the 1980s: 'cervical dystonia'. In 1985 the Canadian Tsui reported for the first time the successful use of botulinum toxin injections in 12 patients,89 which eventually became the standard treatment. During the past few decades hardly any reports on psychogenic cervical dystonia cases have been published.

### Table 2.3. Seven reasons why focal dystonias were regarded as psychogenic

<p>| | |</p>
<table>
<thead>
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<tbody>
<tr>
<td>1</td>
<td>The bizarre nature of the dyskinesias</td>
</tr>
<tr>
<td>2</td>
<td>Their appearance frequently only on certain actions, other motor acts employing the same muscles being carried out normally</td>
</tr>
<tr>
<td>3</td>
<td>Their relief by certain inexplicable trick actions</td>
</tr>
<tr>
<td>4</td>
<td>Their exquisite sensitivity to social and mental stress</td>
</tr>
<tr>
<td>5</td>
<td>The failure so far to find any anatomical, physiological, or biochemical abnormality in any of these conditions</td>
</tr>
<tr>
<td>6</td>
<td>The belief that such patients show overt psychiatric disturbance</td>
</tr>
<tr>
<td>7</td>
<td>A psychopathological interpretation of the significance of, for example, eye closure or neck turning</td>
</tr>
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**Writer's cramp**

In 1713 the Italian physician Bernardino Ramazzini (1633-1714) recognised intense fatigue of the hand and arm, which resulted in failure of power, as an occupational disorder in professional writers.91 In 1844 (published posthumously), Bell most probably described writer's cramp when he reported on an ambiguous condition in which writing had become impossible while the arm strength remained normal.60 In 1861 Duchenne reported on 'crampe des écrivains' in which electricity was not a very successful therapy. However, he
advised an ingenious prosthesis.\textsuperscript{62} He preferred the names 'spasme fonctionnel' and 'paralysie musculaire fonctionelle' because the disorder was not restricted to cramps and could be provoked not only by writing but also by other manual actions.

En résumé, les faits et les considérations exposés précédemment démontrent, comme je l'ai dit au commencement de cette note, qu'il existe une maladie caractérisée par un spasme douloureux ou indolent (contracture, contractions clonique, tremblements), ou par une paralysie musculaire; que ces troubles se manifestent seulement pendant l'exercice de certains mouvements volontaires ou instinctifs; enfin, qu'ils peuvent siéger dans des régions fort diverses.\textsuperscript{62} [In summary, the former findings and considerations show, as I described in the beginning of this report, that there is a disease which is characterised by painful or painless spasms (contracture, jerks, tremor) or paralysis in which the signs only occur during certain (in-)voluntary actions; the involved body parts are diverse.]

Similar to the situation in cervical dystonia, the debate on aetiology started in the early 20\textsuperscript{th} century. In 1914 the German Fritz Mohr (1874-1957) summarised the two conflicting theories in Lewandowsky's \textit{Handbuch der Neurologie}.\textsuperscript{12} Writer's cramp was explained by some authors as a purely organic disorder, e.g. as a reflex cramp through motor nerves that was initiated by painful sensory input. The German physician Moritz Romberg (1795-1873) was mentioned as one of the early advocates (with reference to the 1853 edition of Romberg's \textit{Lehrbuch der Nervenkrankheiten des Menschen}).\textsuperscript{92} Others, including Mohr himself, believed that only people with certain personality characteristics were prone to develop the disorder, a psychological factor possibly being involved. From that view, accurate psychoanalysis would be the best therapy for patients with writer's cramp. Kinnier Wilson's (1878-1937) 1940 edition of \textit{Neurology}, described 'writers' cramp' as an occupational neurosis, physiologically akin to hysteria, and assumed a cortical dysfunction.\textsuperscript{11} Prevention by excluding people prone to develop 'spasms' from certain occupations, was considered the best treatment.

As in other focal dystonias, Marsden advocated the organic nature of writer's cramp (\textit{vide supra}),\textsuperscript{3} which he and Sheehy further demonstrated in a report on 29 patients, (1982).\textsuperscript{93} However, in 1983 Cottraux (France) \textit{et al.} reported on the success of behavioural therapy and biofeedback in 9 of 15 patients with writer's cramp,\textsuperscript{94} and the 1985 edition of John Walton's \textit{Brain's Diseases of the nervous system} stated:
I find the conclusions of Sheehy and Marsden inherently implausible and unacceptable. In my experience even subtle physical signs are absent in the many 'simple' cases that I have seen and neither focal dystonia nor any other organic disorder could in my view impair movements only when they take part in one co-ordinated act while leaving totally unaffected all other precise and complex voluntary actions involving the affected member.\textsuperscript{95}

The 1993 edition stated that writer’s cramp had "in the past been attributed to psychological factors, but there is now good evidence that this is not so". Interestingly, the author referred to the same single publication of Sheehy and Marsden.\textsuperscript{93,96} In 1991 Rivest \textit{et al.} reported for the first time on the use of botulinum toxin for writer’s cramp,\textsuperscript{97} which is currently considered the most effective treatment.

\textbf{Fixed dystonia related to complex regional pain syndrome}

In 1864 Silas Weir Mitchell (1829-1914) described a series of American Civil War (1861-1865) victims with gunshot wounds who developed burning pain and a shiny red skin after nerve injury.\textsuperscript{98,99} He suspected that traumatic nerve irritation was the cause and named the condition 'causalgia'. He recognised that patients might come into an unendurably painful hyperaesthetic state. In 1892, Charcot demonstrated another entity in two patients: 'œdème bleu des hystériques', a painful condition with oedema and blue discoloration of the skin, which may occur in combination with an hysterical limb contracture or paralysis.\textsuperscript{100}
hyperesthesia) or movement disturbances (paralysis and contractures). Mostly, patients were extraordinary persons having characteristics which may be considered hysterical.]

In 1946, Evans renamed the latter disorder 'reflex sympathetic dystrophy', because he suspected involvement of spinal reflexes as well as sympathetic efferent fibres.\textsuperscript{101} It was different from causalgia in that it occurred in the absence of major nerve trauma. However, in 1994, the International Association for the Study of Pain introduced the name CRPS for both conditions: type 1 (reflex sympathetic dystrophy) and type 2 (causalgia).\textsuperscript{102} The diagnostic criteria for CRPS type 1 were: (i) presence of an initiating noxious event, or a cause of immobilization (not obligatory item); (ii) continuing pain, alldynia, or hyperalgesia with which the pain is disproportionate to any inciting event; (iii) evidence at some time of oedema, changes in skin blood flow, or abnormal sudomotor activity in the region of the pain; and (iv) no other condition that would account for the degree of pain and dysfunction. CRPS type 2 has the same characteristics, but is accompanied by nerve injury.\textsuperscript{102}

In 1984 Marsden et al. reported on four 'reflex sympathetic dystrophy' patients who had dystonia, characterised by fixed, predominantly flexion, postures.\textsuperscript{103} They believed it to be 'a distinct clinical syndrome'. Six years later Schwartzman et al. reported on motor disturbances in 43 patients with 'reflex sympathetic dystrophy', in whom the most dramatic characteristic was a dystonic posture in all patients.\textsuperscript{104} The authors hypothesised on a spinal cause. In 1993, a series of 18 patients with similar characteristics was reported.\textsuperscript{105} However, it was remarkable that many patients met the criteria for psychogenic dystonia from 1988 \textit{(vide supra)} (Table 2.1). They concluded that the aetiology of this disorder, psychogenic or organic, was unknown.\textsuperscript{105} In 2004 it was reported that many patients with features of CRPS and dystonia also had features of psychogenic dystonia.\textsuperscript{106} In the same year it was stated that a very large proportion had a primary psychogenic disorder.\textsuperscript{107} In a more recent paper on 110 CRPS type 1 patients with dystonia predominantly characterised by tonic flexion postures, the authors hypothesised that maladaptive plasticity with disinhibition of spinal mechanisms might be the cause.\textsuperscript{108}

Discussion

There has been a continuous vacillation between psychogenic and organic explanations for (i) primary generalised dystonia; (ii) cervical dystonia; (iii) writer's cramp; and (iv) CRPS-
related fixed dystonia. Although at first sight the attributions of the terms psychogenic and organic in Table 2.2 seem quite obvious, it seems more realistic to assume a spectrum with two ends between which attributions were moving. Moreover, the discussion between an organic and psychogenic aetiology has not always been explicit (particularly in the 19th century). The opinions of several authors could only be derived or interpreted from their hypotheses on aetiology and their therapies.

An example is Schwalbe's description of hysterical symptoms in siblings with generalised dystonia. In the late 19th century, Charcot considered hysteria a neurosis, similar to paralysis agitans, epilepsy and chorea, which were diseases without known pathology. For paralysis agitans he expected that the lesion would be discovered. Hysteria appeared a more difficult obstacle for Charcot's clinical-anatomic method and, when describing male traumatic neurosis, he moved towards a psychological conception of hysteria. This was further elaborated by Freud and his followers. Hysteria evolved from a disease in which an organic pathophysiology was suspected but not found, to a psychogenic disease in the late 19th and early 20th century. Recent functional imaging studies in these patients have shown specific cerebral abnormalities. From these studies, it is suspected that affective or stress-related factors modulate cerebral sensorimotor representations through interactions between limbic and sensorimotor networks. It is hypothesised that primitive reflexive mechanisms of protection and alertness, which are partly independent of conscious control, are involved.

Primary generalised dystonia
The patients of Oppenheim made him move to the organic end of the spectrum, whereas Freud and his followers in psychoanalysis, moved in an opposite direction. The improved description of the 'clinical entity' in the 1940s (Herz), the new hereditary cases described in the 1950s (Zeman), and the limited efficacy of psychotherapy in torsion dystonia, as well as the effects of surgical treatments and the lesion studies in the 1960s (Eldridge, Cooper, and Denny-Brown respectively) pushed the explanatory ideas back into the organic again. This culminated in Eldridge's & Fahn's 1975 statement (published in 1976). However, a new movement towards psychogenesis soon followed with the recognition of psychogenic dystonia. Meanwhile, the remaining dystonias kept their position on the organic side of the spectrum, not in the least because of the discovery of the DYT1 gene. Nevertheless, one cannot be too rigid because dystonic disorders with a genetic origin can be triggered by emotional stress.
Cervical dystonia
Bell and Duchenne probably assumed an organic cause for cervical dystonia (Table 2.2). Not much later Charcot, and certainly his student Brissaud, moved to the psychogenic view, in which the interpretation of observing the ‘geste antagoniste’ played an important role. At the time, such terms as ‘torticollis mental’ and ‘torticollis hystericus’ were used on both sides of the Atlantic and dealt with likewise. Psychological and surgical treatments were applied simultaneously in different patients at different places around the 1940s. After Marsden's 1975 presentation, the aetiological ideas on cervical dystonia clearly moved away from the psychogenic to the organic side of the spectrum.

Writer's cramp
To explain writer's cramp, Ramazzini used such terms as ‘fatigue’ and ‘failure of power’. These should be interpreted in the humoral pathophysiological concepts of the time, i.e. animal spirits that flow through the nerves with less power than usual. One would be inclined to consider an organic aetiology here; however, we may question whether Ramazzini was concerned with this question at all. From Bell's description a century later and also from Romberg's work, an organic viewpoint may be recognised. Duchenne again used the term ‘functional’, which, however, does not necessarily imply that he meant a psychogenic aetiology. A clearer distinction came about in the early 20th century, when Mohr mentioned personality characteristics and a psychological factor, and suggested psychoanalysis for treatment. An interesting position was taken by Kinnier Wilson, assuming cortical dysfunction but comparing it to hysteria. Once more, Marsden's 1975 presentation pushed the aetiology of writer's cramp toward the organic side, with a few exceptions in the 1980s.

Fixed dystonia related to complex regional pain syndrome
Charcot's demonstration of two patients with 'oedème bleu des hystériques' occurred in a period in which he was moving towards a psychological explanation of hysteria. Marsden et al. expressed the opinion that the similarities between CRPS cases with dystonia over the world suggested its existence as a distinct clinical syndrome.\(^\text{103}\) In contrast, Sa et al. stressed that most cases satisfied the criteria for psychogenic dystonia, and should, therefore, be considered as such.\(^\text{107}\) But these are based on expert opinion. Such statements are not like a gold standard and should, therefore, be used with caution. Moreover, it is remarkable that the reasons why CRPS-related fixed dystonia is considered psychogenic are at least partly the same as the arguments that were used in the past to
explain why focal dystonia was psychogenic (Table 2.3): (i) the dystonia in CRPS may be considered incongruent with classical dystonia; (ii) may be inconsistent over time; (iii) weakness, described in the majority of CRPS cases, might be interpreted as false; (iv) sensory abnormalities, which fit the diagnosis of CRPS, might be interpreted as false sensory findings; and (v) sometimes, psychiatric abnormalities are present. In recent times significant motor cortex abnormalities were found in CRPS.\textsuperscript{114,115}

It is clear that the discussions on the psychogenic or organic aetiology of dystonia have been emotional. In some of the periods, particularly during the 20\textsuperscript{th} century, strong believers as well as non-believers may be recognised. Charcot isolated hysterical disorders from other neurologic diseases. In his view, environmental factors (‘agents provocateurs’) were involved in its pathogenesis. The rise of the psychoanalytic movement, following the work of Freud at the beginning of the 20\textsuperscript{th} century, caused important disagreements between supporters of organic and psychogenic explanations. This was not specific to the interpretation of dystonia, but more generally reflected the division between biologically and psychoanalytic oriented neuropsychiatrists at the time. The success and popularity of psychoanalysis, as well as the lack of an organic substrate for dystonia, encouraged psychogenic theories. As the 20\textsuperscript{th} century proceeded, knowledge in favour of a somatic origin of early-onset generalised dystonia accumulated. Marsden, a leading neurologist in movement disorders, convinced the neurological community in the 1970s and 1980s that both generalised and focal dystonia were somatic entities. However, psychogenic dystonia re-emerged, but as a special category. Nowadays, psychogenic dystonia is thought to be “common” in specialised movement disorders clinics.\textsuperscript{116}

The recognition of the hereditary character of dystonia played an important role in attributing an organic nature in the first as well as the last decades of the 20\textsuperscript{th} century. If dystonia had existed as an entity and its hereditary character had been recognised previously, it would probably have been interpreted in a different way, because of the particular concepts of the late 19\textsuperscript{th} century. In this period several neuroses were considered hereditary, in fact a favourite subject in the interpretation models of Charcot.\textsuperscript{117} Similar to contemporary psychiatrists (the French Benedict-Augustin Morel (1809-1873) and Valentin Magnan (1835-1916)), he assumed that degeneration was a constitutional factor in certain families (‘neuropathic families’) in which neuroses including hysteria, alcoholism, and epilepsy could be transformed during the passage from one generation to the next. Hysteria in a parent could be inherited as epilepsy in the child.\textsuperscript{109,118} In the 20\textsuperscript{th} century, following delineation of dystonia as an entity and following
new discoveries in genetics, the hereditary character led to new insights. Today it is recognised that more than 14 genes are implicated in different monogenic dystonia syndromes, which are frequently inherited as autosomal dominant conditions with reduced penetrance. Most cases of early-onset torsion dystonia are associated with the DYT1 gene mutation. Familial occurrence of cervical dystonia or writer's cramp has been described but appears to be rare.

Medical problems nearly always unravel because of the advent of a new technology, skill, or understanding of a hitherto unknown system of disease. One example is the unravelling of the electric nature of nerve action in the 18th and 19th century. Ideas on whether or not animal electricity existed and played a role in the nature of nerve conduction, were put forward at the end of the 18th century by Galvani and denied by Volta. The confirmation had to await more sophisticated sensitive measuring devices such as the galvanometer invented by Du Bois-Reymond in the 1840s, after which observation of the action potential became possible. What will be the future 'sophisticated sensitive measuring device' that will finally lead to the understanding of dystonia? We believe that the increasing knowledge resulting from neurophysiological and imaging studies, combined with genetic methods, will provide the insight that the explanation of dystonia cannot just be interpreted in terms of organic or psychogenic. These modern methods may show that the interaction of genetic and environmental factors is more complex than was previously thought. When reviewing the pathophysiology of primary adult-onset focal dystonia, Defazio et al. suggest that in human focal dystonia there may be an overload of a predisposed sensory system resulting from peripheral injury or repetitive motor activity in a certain part of the body, or both, causing sensory receptive changes in the corresponding cortical brain areas and leading to abnormal regulation of inhibitory interneuronal mechanisms at brainstem or spinal cord level. There seems to be an abnormality of sensorimotor integration and cortical excitability beyond the symptomatic body part. In both generalised and focal dystonia neurophysiological and functional imaging studies indeed point towards abnormalities in the sensorimotor circuitry, which result in a vulnerable central nervous system. Some of these phenomena have been found in asymptomatic gene carriers, as well as in representations of unaffected body parts. It is suspected that a 'second hit' is needed to bring the central nervous system out of balance, which leads to dystonia. Musician’s dystonia is an interesting example. In a transcranial magnetic stimulation study, cortical changes were found in musicians compared to healthy controls, and these changes were
more marked in those with musician’s dystonia. It is hypothesised that musician’s dystonia is a form of training-induced dystonia.\textsuperscript{122}

These suggestions, in particular the assumption of abnormal regulation of inhibitory interneuronal mechanisms at brainstem or spinal cord level, bring us back to Sherringtonian neurophysiology as already suggested by Denny-Brown in the 1960s, when he found that damage in different anatomical structures could have the same consequence, pointing to a basic neurophysiological principle, the final common path, that had been conceived around the turn of the 19\textsuperscript{th} to the 20\textsuperscript{th} century by his teacher Charles Scott Sherrington (1857-1952).\textsuperscript{123,124} This may still be a valid explanation of the phenomenology, if not the underlying causation of dystonia in modern terms.

Today, psychogenic dystonia is considered a disorder that results from an underlying psychiatric illness. Its diagnostic criteria have remained unchanged for decades (Table 2.1). In the meantime, however, the border between neurology and psychiatry has been less well defined. For example, schizophrenia,\textsuperscript{125} autism,\textsuperscript{126} and primary dystonia\textsuperscript{113} are now considered neurofunctional disorders. Additionally, it has been shown recently that patients with cervical dystonia or blepharospasm have distinct neuropsychiatric and personality profiles of the anxiety spectrum.\textsuperscript{127} Another study shows high psychiatric comorbidity in cervical dystonia, which is unlikely to be a mere consequence of chronic disease and disfigurement.\textsuperscript{128} It is attractive to see psychogenic disorders as the consequence of functional crashes in anatomically normal brains. In these disorders, abnormalities found in neurophysiological and functional imaging studies may be interpreted as signs of organic dysfunction. We have only traced two transcranial magnetic stimulation studies on psychogenic dystonia. Interestingly, one of these found similar abnormalities in both organic and psychogenic dystonia: reduced short and long-interval intracortical inhibition and cortical silent period, and an increased cutaneous silent period.\textsuperscript{116} The other detected difference: patients with organic dystonia had an increased response to paired associative stimulation compared to patients with psychogenic dystonia.\textsuperscript{129} The authors of the latter study concluded that abnormal plasticity is a hallmark of organic dystonia in contrast to psychogenic dystonia.

If we hypothesise further, assuming abnormal regulation of inhibitory interneuronal mechanisms as mentioned above, neurophysiological and functional imaging studies may help to explain dystonia in CRPS due to peripheral injury leading to similar sensory receptive changes. Such mechanisms may also be in play in dissociation disorders, including conversion disorder,\textsuperscript{111,112,130-132} thereby associating primary dystonia, CRPS-related fixed dystonia, and sensory and motor disorders in conversion disorder.
Conclusions

Opinions on whether dystonia is either organic or psychogenic continuously changed on a spectrum between the two extremes over the described period. Genetic studies, the limited efficacy of psychotherapy, the effects of surgical treatments, lesion studies, and the recognition that focal dystonias may be minor forms of generalised dystonia pushed the explanatory ideas in the direction of organic. We have seen how insights were influenced by contemporary general pathophysiological concepts (humoral pathophysiology in the pre-1800 period, solid pathophysiology reflected by the clinical-anatomical method thereafter, psychological pathophysiology after about 1900, and genetic and molecular pathophysiology in recent decades), as well as by various research methods, from which we have learn to be prudent with the interpretation of results and to reflect on epistemological mechanisms. Nevertheless, with these reservations in mind, modern neurophysiological and imaging studies may open new ways for the interpretation of dystonia. In both generalised and focal dystonia, studies point towards abnormalities in the sensorimotor circuitry, resulting in a vulnerable central nervous system. They indicate that the old distinction between psychogenic and organic is not easily applicable and perhaps should be abandoned. Similar mechanisms may be in play in CRPS-related fixed dystonia and sensory and motor disorders in conversion disorder. Hypotheses made on the basis of neurophysiological and functional imaging studies need further testing in these groups of patients. In addition, genetic studies may provide further insight. Until more knowledge is available, we must keep in mind the lessons from history and remember 1975:

In the past, many victims of dystonia and their families have been caused anguish and hardship over and above that caused by the disease itself owing to the frequent misdiagnosis of the symptoms as manifestations of a psychiatric ailment.  

Once hurt, twice shy.
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